UNIVERSITY of TASMANIA

Identification of Executive Function Difficulties in Preterm Preschool and Kindergarten Children

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Declaration of Originality

This thesis contains no material which has been accepted for a degree or diploma by the University or any other institution, except by way of background information and duly acknowledged in the thesis, and to the best of my knowledge and belief no material previously published or written by another person except where due acknowledgement is made in the text of the thesis, nor does the thesis contain any material that infringes copyright.

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Statement Regarding Published Work Contained in this Thesis

Statement of Ethical Conduct

The research associated with this thesis abides by the International and Australian codes on human and animal experimentation, the guidelines by the Australian Government's Office of the Gene Technology Regulator and the rulings of the Safety, Ethics and Institutional Biosafety Committees of the University.

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Abstract

Background: It is known that individuals born preterm are at risk of developing executive function and behavioural difficulties. Nevertheless, most studies in this field have focussed on school-age children, as investigation of executive functions of younger preschool and early school-age children (under six years old) has only recently gained attention. Consequently, there is little knowledge about social and perinatal risk factors contributing to executive function difficulties in young preterm children and how to best identify those at risk of developing executive function difficulties. The overall aims of the present thesis were to explore the relationship between executive functioning and social and perinatal risk factors in four- to five-year-old preterm children, and how to best identify children most at risk of executive function difficulties prior to starting formal schooling.

This thesis integrates three published or submitted articles, each addressing a separate research question related to the overarching thesis aims. All articles incorporate information regarding a sample of 141 children born preterm (< 33 weeks of gestation) and 77 term comparison children who were assessed using standardized measures of general intelligence and performance-based executive function tests prior to starting kindergarten at the age of four years. Parental and teacher reports of executive functioning and behaviour were completed when the children commenced kindergarten at four to five years of age.

Study 1

Aim: To establish whether four- to five-year-old preterm children had more executive function difficulties than their term peers, and if so, to identify the social and perinatal risk factors associated with such discrepancy.

Methods: The preterm and term groups were compared on measures of intelligence and executive functions using independent group *t*-tests, and multivariate regression analyses were performed to identify social and medical risk factors predictive of intelligence and executive functioning in the preterm group.

Results: The preterm group performed significantly more poorly than the term group on all intelligence and performance-based executive function assessments. The parental reports relating to executive function for preterm and term children did not significantly differ, but the teachers reported more executive function difficulties for the preterm group than the term group. Overall, higher social risk, and in particular lower educational level of the main caregiver, was the strongest predictor for the preterm children's intelligence and executive function difficulties.

Study 2

Aim: To determine whether specific performance-based executive function assessment tools were associated with executive functioning in everyday life as reported by parents and teachers of four- to five-year-old preterm and term children.

Methods: The associations between performance-based intelligence, executive function assessments, and executive function questionnaires were explored by using multiple regression analyses.

Results: Performance-based intelligence and executive function assessment results did not have strong associations with the reported executive function difficulties.

Study 3

Aim: To investigate the congruency of parent and teacher reporting of executive functions and behaviour of kindergarten-age preterm and term children.

Methods: Parent and teacher reporting of executive function and behaviour were compared using 2x2 mixed ANOVA and Kappa statistics.

Results: Parents reported higher levels of executive function and behavioural difficulties than the teachers for both groups combined (preterm and term). The parent and teacher reports of behaviour and executive function differed significantly and were not in agreement relating to which children exhibited clinically significant executive function and behavioural problems.

Conclusion*:* Formal guidelines relating to surveillance of young preterm children's executive function and behaviour difficulties are needed, especially relating to selection criteria for follow-up (i.e., based solely on gestational age or also other factors), recommended assessment batteries, and the timing of the assessments. To assist in achieving these outcomes, it is recommended that future studies focus on improving the ecological validity of psychometric assessments of preterm children. Increased accuracy in early identification of at-risk preterm children could enable timely executive function and behavioural support and intervention, which could lead to narrowing the current gap in developmental and educational outcomes between preterm and term children.

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Publications

Publications directly arising from the work described in this thesis:
O'Meagher, S., Kemp, N., Norris, K., Anderson, P., & Skilbeck, C. (2017). Risk factors for executive function difficulties in preschool and early school-age preterm children. *Acta Paediatrica, 106,* 9, 1468-1473.

O'Meagher, S., Norris, K., Kemp, N., & Anderson, P. (2018). Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice. *Child Neuropsychology*. (Advance online publication).

O'Meagher, S., Norris, K., Kemp, N., & Anderson, P. Parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten. Submitted to *Applied Neuropsychology: Child.* (In press).

Conference presentations using the work from the current project:
O'Meagher, S., Norris, K. & Kemp, N. (2017, November). *Cognitive outcomes and intervention attendance of Tasmanian preterm preschoolers.* Poster presentation at the Allied Health Symposium, Hobart, Australia.

O'Meagher, S., Kemp, N., Norris, K., & Anderson, P. (2017, September). *Preterm preschoolers' performance on Day-Night and Shape School executive function tasks.* Poster presentation at the 6th Scientific Conference of the Federation of the European Societies of Neuropsychology (FESN), Maastricht, the Netherlands.

O'Meagher, S., Kemp, N., Norris, K., & Anderson, P. (2017, July). Sex differences in executive functions of preterm and term preschool children. *Journal of the International Neuropsychological Society* (2017), 23, Suppl. 2. Oral presentation at the International Neuropsychological Society Midyear Conference, Cape Town, South Africa.

O'Meagher, S., Kemp, N., Skilbeck, C. & Anderson, P. (2015, July). *Does the performance of preterm preschool children on executive function tests predict executive functioning in real life?.* Poster presentation at the 5th International Neuropsychological Society Pacific Rim Conference, Sydney, Australia.

Statement of Co-Authorship

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Article 1 Risk factors for executive function difficulties in preschool and early school-age preterm children Located in Chapter 2

Candidate was the primary author responsible for the idea, data collection, data analysis and main manuscript preparation (70%), authors 1 and 2 contributed to the refinement and presentation (10% each), and authors 3 and 4 contributed to the idea and refinement (5% each).

Article 2 Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice Located in Chapter 3

Candidate was the primary author responsible for the idea, data collection, data analysis and main manuscript preparation (70%), and authors 1 and 2 contributed to development, refinement and presentation (15% and 10% respectively). Author 3 contributed to refinement (5%).

Article 3 Parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten Located in Chapter 4

Candidate was the primary author responsible for the idea, data collection, data analysis and main manuscript preparation (70%), and authors 1 and 2 contributed to development, refinement and presentation (15% and 10% respectively). Author 3 contributed to refinement (5%).

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Abbreviations

ASEBA Achenbach System of Empirically Based Assessment

BRIEF-P Behavior Rating Inventory of Executive Function–Preschool Version

EF executive function

IQ intelligence quotient

NEPSY-II Developmental Neuropsychological Assessment Battery, 2nd edition

RHH Royal Hobart Hospital

WPPSI-III Wechsler Preschool and Primary Scale of Intelligence, 3rd edition

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Chapter 1. Introduction

Introduction

The current thesis presents three papers looking at how to best identify four- to five-year-old preterm children at risk of developing executive function and behavioural difficulties. “Executive functions” is an umbrella term for higher order cognitive processes, which progressively develop during childhood and adolescence (Hughes, Ensor, Wilson, & Graham, 2010). Preterm birth refers to a birth occurring before 37 weeks’ gestation (Goldenberg, Culhane, Iams, & Romero, 2008). The term “extremely preterm” is used for gestation < 28 weeks’, “very preterm” for <32 weeks’ and “moderate preterm” for 32–33 weeks’ gestation (Goldenberg et al., 2008).

Executive Functions and Their Development

In 1991, Welsh, Pennington, and Groisser presented evidence for at least three executive factors: working memory and planning, inhibition of maladaptive prepotent responses, and attentional flexibility. Similar concepts of working memory, inhibition and mental set shifting, have also been named as main executive function components by more recent researchers (Lehto, Juujärvi, Kooistra, & Pulkkinen, 2003; Miyake, Friedman, Emerson, Witzki, & Howerter, 2000). While other executive function structures have been suggested by researchers like Lezak (1995) and Barkley (2012), the three-component model of executive functions (mental set shifting, working memory and inhibition) has gained most support. Shifting refers to the ability to change between mental sets and tasks, working memory consists of holding and manipulating multiple pieces of information in the mind, and inhibition enables overriding of more dominant or prepotent responses (Lehto et al., 2003; Miyake et al., 2000). The three components have been found to be partially independent but still intercorrelated in adults and adolescents (Lehto et al., 2003; Miyake et al., 2000).Recently, Miyake and Friedman (2012) have revised their model of executive functioning to consist a common executive function factor which includes inhibition and executive attention and separate shifting and updating (working memory) factors. More complex executive function tasks, such as planning and organizational skills and problem solving, utilize a combination of above processes (Diamond, 2013). In children seven years and older, executive functions have been found to be separable into these three components (Huizinga, Dolan, & van der Molen, 2006). However, some researchers have suggested that executive functions have a more unitary construct in the preschool years (Hughes et al., 2010; Wiebe, Espy, & Charak, 2008; Wiebe et al., 2011). Thus, it has been recommended that when adopting a modular framework of executive functions, especially in children, caution should be exercised, as executive processes could be better viewed as functional networks constructed over the course of development (Bernstein & Waber, 2007).

Executive functions have also been suggested to have both “hot” (affective) and “cool” (cognitive) components. Hot and cool executive functions have different relationships with each other and with general intellectual function (Hongwanishkul, Happaney, Lee, & Zelazo, 2005; Zelazo & Carlson, 2012). Hot executive functions refer to social and emotional domains, and include mood, self-awareness, emotional control, moral judgement and social information processing (Hongwanishkul et al., 2005; Zelazo & Carlson, 2012). Cool executive functions consist of cognitive processes, such as attention, working memory, planning, and monitoring performance (Hongwanishkul et al., 2005; Zelazo & Carlson, 2012). There is growing evidence that hot and cool processes are separate, yet correlated, functions. Cool executive functions are more strongly associated with cognitive problem solving and academic achievement and hot regulation with behaviour and attention control difficulties in children (Brock, Rimm-Kaufman, Nathanson, & Grimm, 2009; Hongwanishkul et al., 2005; Thorell, 2007; Willoughby, Kupersmidt, Voegler-Lee, & Bryant, 2011). It has been suggested that the hot and cool executive functions are controlled by different areas of the brain, with hot executive functions being associated with the orbitofrontal areas and cool executive functions by the dorsolateral prefrontal areas (Anderson, Anderson, Northam, Jacobs, & Mikiewicz, 2002; Payne, Hyman, Shores, & North, 2011).While hot and cold executive functions can overlap in an individual, it is possible to have hot executive function difficulties in the absence of cool executive function problems. Thus, assessment of both hot and cold executive functions is important when evaluating higher cognitive processes.

The scarcity of neuropsychological tests for preschoolers has limited the extent of research on executive functions in this population (Baron, Kerns, Müller, Ahronovich, & Litman, 2012). Furthermore, until quite recently, preschool children were thought to lack executive functioning (Isquith, Crawford, Espy, & Gioia, 2005). Nevertheless, it has been demonstrated that some executive functions, such as working memory and attentional control, in fact start developing in early infanthood, and as early as at eight to 12 months of age (Garon, Bryson, & Smith, 2008; Diamond, 2006; Senn, Espy & Kaufmann, 2004). Executive functions develop sequentially and are simpler in preschool years than later in development (Hughes et al., 2010; Klenberg, Korkman, & Lahti-Nuuttila, 2001; Wiebe et al., 2011). The development of basic inhibitory functions precedes the development of more complex functions, such as mental set shifting and planning. Attention control and working memory are functional by 12 months and keep developing during school years (Diamond, 1988, 1991). Between the ages of three and five years, children's executive functions undergo fundamental changes (Carlson, 2005; Garon et al., 2008). Capacity to shift thinking emerges at around three to four years and keeps on improving for years, unlike inhibition, which seems to consolidate earlier and improves rapidly between three and five years (Carlson & Moses, 2001; Diamond & Taylor, 1996; Lehto et al., 2003). For example, three-year-old children can inhibit incorrect responses as well as four- to five-year-olds but less efficiently, and five-year-old children are much faster and accurate in tasks requiring shifting than four-year-olds (Espy, 1997). As shifting ability is slower to consolidate, some researchers have even suggested that executive functions are primarily divided into working memory and inhibition until teenage years (Lee, Bull, & Ho, 2013). Nevertheless, as mentioned earlier, differences in shifting ability can be identified between different age groups (for example, four- and five -year-olds), and thus individual differences in shifting ability exist even in preschoolers. Overall, assessment of all executive function components (mental set shifting, working memory and inhibition), and also hot and cool executive functions, is warranted in the pre-school population in order to identify children who may need support for their executive function development. Figure 1 summarises executive function development across the lifespan.

Preterm Birth and Associated Risks and Complications

The number of children surviving preterm birth has increased with the advancements in perinatal care (Institute of Medicine, 2007). It is estimated that in developed countries, such as Australia, 5-10% of all pregnancies end preterm (<37 weeks' gestation), and about 1-2% end before 32 weeks' gestation (Cheong & Doyle, 2012; Purisch & Gyamfi-Bannerman, 2017). Many children born preterm are vulnerable to developmental, neurological and neurosensory difficulties. Over 10% of premature children develop major physical and intellectual disabilities, 50% develop neurobehavioural problems and 40% require special education or assistance in educational settings (Anderson & Doyle, 2004; Mikkola et al., 2005; Taylor, Klein, Drotar, Schluchter, & Hack, 2006). Importantly, even in the absence of major neurological problems, executive function difficulties and other higher level cognitive problems have been detected in up to 50-70% of preterm children who otherwise meet the major developmental milestones, such as walking and talking, within the normal limits (Caravale, Tozzi, Albino, & Vicari, 2005; Msall et al, 1991; Taylor, Klein, & Hack, 2000).

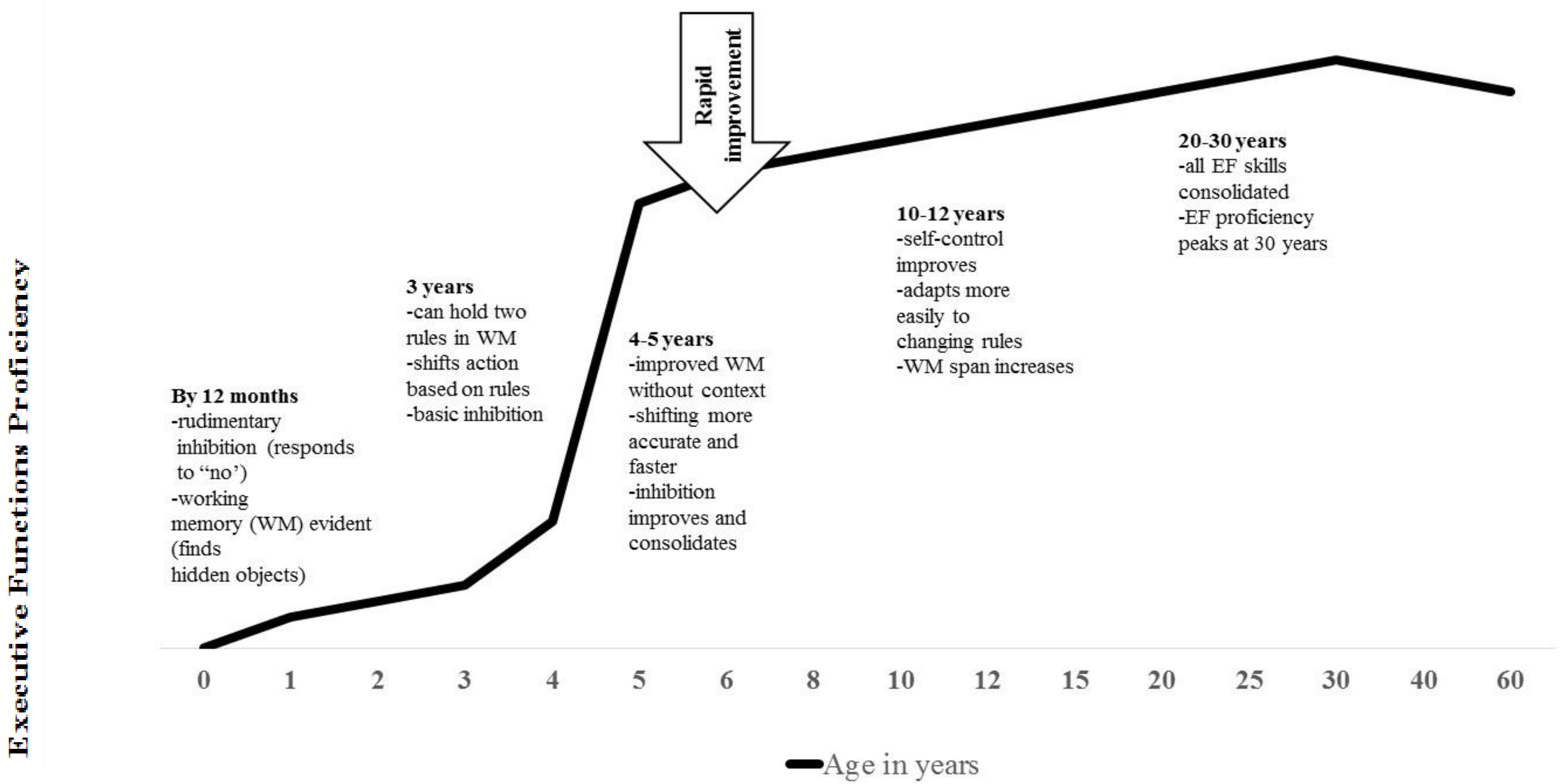


Figure 1. Development of executive function proficiency. Sources: Carlson, 2005; Diamond, 1988, 1991, 2006; Garon et al., 2008; Hughes et al., 2010; Lehto et al., 2003; Weintraub et al. ,2013; Wiebe et al., 2011).

Thus, while some preterm children are spared from major developmental disabilities, they are still at risk of having executive function and behavioural difficulties (Arpi & Ferrari, 2013; Bhutta, Cleves, Casey, Cradock, & Anand, 2002; Delobel-Ayoub et al., 2009).

Executive function difficulties have been identified in preterm children at all levels of prematurity (extremely preterm <28 weeks' gestation, very preterm <32 weeks' gestation and moderately preterm 32-33 weeks' gestation; Mulder, Pitchford, Hagger, & Marlow, 2009). While majority of the research has focussed on extremely and very preterm children, recently studies have revealed that even late preterm children born between 34 to 36 weeks' gestation exhibit executive dysfunction (Baron et al., 2012; Brumbaugh, Hodel, & Thomas, 2014). Preterm children perform significantly more poorly than their peers on EF measures of attention, inhibition and planning as early as eight months of age (de Jong,Verhoeven, & van Baar, 2015; Sun, 2011). At preschool age, deficits in working memory (Baron et al., 2012; Loe, Chatav, & Alduncin, 2015; Rose, Feldman, Jankowski, & Van Rossem, 2011), inhibition (Loe et al., 2015; Orchinik et al., 2011), and mental flexibility (shifting) (Baron, Erickson, Ahronovich, Baker, & Litman, 2011) have been reported. In general, the results have been mixed, with some researchers indicating there are global EF deficits, while others have only found difficulties in specific components of EF, such as working memory or inhibition (Aarnoudse-Moens, Duivenvoorden, Weisglas-Kuperus, Goudoever, & Oosterlaan, 2012; Anderson & Doyle, 2004). It is possible that variation in task demands may explain why researchers have not consistently found similar patterns of executive dysfunction in preterm children (Taylor & Clark, 2016). It appears that preterm children particularly struggle with increasing EF demands in more complex tasks, while they may be able to pass simpler ones (Baron et al., 2012). Furthermore, current EF assessment tools may tap into latent factors, especially at preschool age, as EFs have a more unitary construct (Hughes et al., 2010; Wiebe et al., 2008; Wiebe et al., 2011). Particular EF assessment subtests may measure more than one EF component (Huizinga et al., 2006). For example, an inhibition test may also tap into working memory, or there may be other latent factors, such as processing speed, which impact on EF assessment results (Mulder, Pitchford, & Marlow, 2011).

While there have been many studies of executive functions and behaviour in the older (over six years) preterm population, there have been fewer to focus on preterm children at the beginning of their school life. In the studies that have examined preschool and early school-age preterm children, behavioural and executive function difficulties have been detected (Aarnoudse-Moens, Smidts, Oosterlaan, Duivenvoorden, & Weisglas-Kuperus, 2009; Arpi & Ferrari, 2013; Loe et al., 2015). However, it should be noted that few studies have specifically investigated the four- to five-year-old age group, when children are entering school in some countries, in relation to executive functioning. As mentioned earlier, children's executive functions undergo fundamental changes at that age. It is important to understand how preterm children's executive functions are developing in relation to those of term children at this crucial period. Any executive function difficulties for preterm children should be identified early, in order to target intervention to the preterm children who may be most at need of support. Children's brains have sensitive periods for different cognitive functions, such as language development and visual processing (Lewis & Maurer, 2005; Ruben, 1999), when they are especially malleable. During sensitive periods, such as those from four to five years for executive functions, cognitive functioning could be improved if risk factors (such as deprived social environment) for development were mitigated by providing support and intervention. There are psychosocial, parenting and educational intervention programmes, such as Child FIRST and Head Start, which have been shown to have a positive impact on cognitive and behavioural development and improve executive functioning of children living in high social risk families (Barnett, 1995; Hertzman & Wiens, 1996; Lowell, Carter, Godoy, Paulicin, & Briggs-Gowan, 2011). While similar programs should in theory also have positive outcomes for preterm children, little is known about their long-term efficacy in this population (Spittle, Orton, Anderson, Boyd, & Doyle, 2015).

Being born preterm exposes babies' vulnerable brains to many risks which underlie executive function and behavioural difficulties. Prenatal brain development, in addition to having a rapid rate of growth, is also complex: the differentiation of pre-myelinating oligodendrocytes, growth of microglia, axons, and subplate neurons, expansion of the thalamic nuclei, and increase in cortical surface area and gyral formation in the cerebral cortex are programmed to occur before the birth (Nosarti et al., 2011). A majority of brain development occurs during the third trimester of gestation, with the volume of the whole brain more than doubling, and the volume of cortical grey matter increasing approximately fourfold (Huppi et al., 1998). Being born preterm can have an extensive negative impact on the developing brain due to its immaturity, the intensive care interventions that preterm babies may endure, such as artificial ventilation and surgeries, and medical complications preterm children may suffer, such as infections and brain haemorrhages (Nosarti et al., 2011; Roberts et al., 2008; Stoll et al., 2004). It is known that children sustaining brain insults prior to and around the time of birth are most at risk for global executive function problems than children who acquire brain insults at a later age (Anderson et al., 2010; Mulder, Pitchford, Hagger, & Marlow, 2009). Insults to and/or reduction of white matter have been associated with neuropsychological, learning and behavioural difficulties in the preterm population (Feldman, Lee, Yeatman, & Yeom, 2012; Northam, Liégeois, Chong, Wyatt, & Baldeweg, 2011). The reduced amount of total white matter seems to be strongly related to poorer outcomes, such as lower IQ, academic and behavioural difficulties (Northam, et al., 2011; Taylor et al., 2011; Vollmer et al., 2006). Primary injury to the white matter may also result in secondary abnormalities in cortical development via interruption to axonal connections (Wyatt, 2010). Researchers have demonstrated that preterm children can have altered white matter despite having no significant neonatal brain injury in the neonatal period or medical problems. These white matter abnormalities have been shown to be relevant to executive function difficulties. For example, Woodward, Clark, Pritchard, Anderson and Inder (2011) found that executive function impairments in four-year-old children were confined to preterm children with white matter abnormalities on brain scans.

Risk factors associated with preterm children's executive functions. In the first article of the current thesis we investigated risk factors associated with preterm children's executive functions. Traditionally gestational age and low birth weight have been used as the main determinants for follow-up of preterm children. Many studies have shown the risk of major disability, developmental delays and cognitive problems increases with decreasing gestational age, especially in the extremely preterm population (Bhutta et al., 2002; Kerstjens, de Winter, Bocca-Tjeertes, Bos, & Reijneveld, 2012; Moore, Lemyre, Barrowman, & Daboval, 2013). Nevertheless, some researchers have not found strong associations between gestational age and cognitive outcomes, especially relating to executive functions (Bos & Roze, 2011; Lundequist, Böhm, & Smedler, 2013; Réveillon et al., 2013; Østgård et al., 2016).

Medical complications associated with prematurity, such as hypoxia (Anderson & Doyle, 2006; Newman, DeBastos, Batton, & Raz, 2011), necrotizing enterocolitis (Chou at al., 2010; Lodha, DeBastos, Batton, & Raz, 2010; Mikkola et al., 2005), hypoglycemia (Duvanel, Fawer, Cotting, Hohlfield, & Matthieu, 1999), severe infections (Stoll et al., 2004), and application of postnatal glucocorticoids (Yeh et al., 2004), can also impact preterm children's outcomes. Nevertheless, many of these risk factors are overlapping, and, in fact, it has been reported that some preterm children (up to 40% of them with neurodevelopmental impairments) are at an increased risk of having multiple complications, while others remain relatively free of medical complications (Hirschberger et al., 2018). Overlapping medical complications can make it difficult to examine the impact of an individual medical complication, such as lung disease, on preterm outcomes, as strict exclusion criteria must be incorporated into study designs to control for the overlapping medical risks. An alternative is to use the length of stay at the hospital as a proxy for medical complications, as sicker preterm children with several medical complications tend to have longer hospital stays (Niknajad, Ghojazadeh, Sattarzadeh, Hashemi, & Shahgholi, 2012). Nevertheless, this approach has its limitations, as hospital stay length is only an approximation of medical risks and may be influenced by other factors. Furthermore, there has been mixed evidence regarding how much perinatal complications, such as lack of oxygen, surgeries and brain haemorrhages, impact on the executive functions of preterm children. For example, Duvall, Erickson, and Lowe (2015) found that perinatal medical risks negatively predicted performance on inhibition, working memory, and cognitive flexibility in preschool children (such that higher levels of risk resulted in reduced executive function performance); however, Lundequist and colleagues (2013) did not observe such clear links with perinatal complications and executive functioning in preterm preschool children.

Overall, medical and birth factors, such gestational age and birth weight, have been shown to impact the major outcomes of preterm children, such as cerebral palsy and intellectual disability, but less is known how much they affect higher level cognitive processes, such as executive functions and behaviour, especially during the period between infancy and school-age when many preterm children are not routinely monitored. The role of other factors, such as the sex of the child and social factors (e.g., parental education level and work status), may also contribute to the executive function outcomes. Studies looking at combination of these social and medical risk factors are scarce, especially when it comes to higher order cognitive and behavioural processes in young preschool and early school-age children. It is necessary to understand which preterm children are most at risk of developing executive function and behavioural issues, in order to provide them with support and intervention as early as possible. The evidence about sex differences in executive functions of typically developing children has been mixed, with some researchers indicating that boys have more executive function difficulties (Klenberg et al., 2001; Raaijmakers et al., 2008), but others not finding such differences (Wiebe et al., 2011). While the impact of sex on executive functions has been not been widely examined in the preterm population, it is known that male preterm individuals have poorer general outcomes, such as lower IQ and language skills, than preterm females (Johnson et al., 2009; Marlow, Wolke, Bracewell, Samara, & the EPIPAGE Study Group, 2005). While behavioural problems are generally more common in the male population, it appears that preterm boys have a disproportionately higher prevalence of behavioural difficulties compared to preterm girls (Reijneveld et al., 2006; Samara, Marlow, & Wolke, 2008). Very little is known about the impact of sex on executive functions of preterm subjects. O'Meagher, Norris, Kemp and Anderson (2017) found that preterm girls outperformed preterm males on some executive function tasks (naming, inhibition, and switching), although these sex differences were no greater than those observed in the term group. In a study by Urben et al. (2017), it was reported that executive function differences between boys and girls was limited to those of born at 27 weeks or earlier. The six-year-old boys had poorer executive control than did girls, but this difference was not as notable in children born after 27 weeks' gestation. Possible sex differences in preterm population require further investigation across different gestational ages, different types of executive function tasks, and age groups.

Social risk and executive functions. Studies of general populations have indicated that children growing up in high social risk families can have more behavioural and executive function problems than children from families with lower social risk (Hackman & Farah, 2009; Merz & McCall, 2011; Msall, Avery, Msall, & Hogan, 2007; Palfrey, 2006). High social risk families can be characterized in this context by factors such as low parental education level, parental unemployment, low maternal age at the birth of the child, and single parenthood (Ardila, Rosselli, Matute, & Guajardo, 2005; Cserjesi et al., 2012; Sarsour et al., 2011; Voss, Jungmann, Wachtendorf, & Neubauer, 2012). It appears that social environment has a greater impact on cognitive outcomes than genetics (Turkheimer, Haley, Waldrom, D'Onofrio, & Gottesman, 2003). The negative impact of a higher social risk on development, behaviour and emotional wellbeing and academic achievement has also been observed in the preterm population (Luu, Ment, Allan, Schneider, & Vohr, 2011; Potijk, Kerstjens, Bos, Reijneveld, & de Winter, 2013; Potijk, Winter, Bos, Kerstjens, & Reijneveld, 2014). Interestingly, while medical factors can cause more severe cognitive deficiencies, social factors can predict both mild and severe cognitive deficits in preterm children (Beaino et al., 2010). In summary, the social environment has been shown to impact cognitive development of school-age preterm children while lower gestational age and medical complications may cause more severe disabilities. Nevertheless, little is known about how these factors interact at preschool age, and which of them are the best predictors of executive dysfunction at that age. While gestational age and medical complications cannot be ameliorated, social risk factors can be addressed. This provides an important avenue for targeted intervention to improve the outcomes of preterm children.

Executive functioning in preterm children. With the advancement in neonatal care of preterm babies, the numbers of preterm children with major disabilities are declining. There were significantly fewer severe developmental delays (3.7% vs.7.3%) and severe disability (3.7% vs.7.8%) in preterm children born in 2005 than those born in 1997 (Doyle, Roberts, Anderson, & the Victorian Infant Collaborative Study Group, 2010). In contrast, it has been recently reported that preterm children born from the year 2000 onwards are at a higher risk of developing executive dysfunction, especially in working memory and planning and organizational skills, than those born in the 1990s (Burnett et al., 2018). Thus, in consideration of potential remediation, surveillance of the development of executive functions of preterm children is clearly justified, even in the absence of major disabilities. Given the limited government health and educational resources for children's assessment and support services, this raises a question: which preterm children need follow-up relating to their executive function development? Should lower gestational age be used as the only criterion to guide follow-up of preterm children, or should other possible risk factors, such as the sex of the child, medical factors and even social risk be considered when selecting children for follow up? It is still unclear how the above factors are associated with development of executive functions of young preterm children. These issues are investigated in the first paper of this thesis.

Assessment of Executive Functions

Performance-based executive function assessment and questionnaires. Discrepancies between performance-based executive function assessment tools and measures of everyday life functioning, such as executive function questionnaires, have been reported by several researchers in general paediatric and adult populations (Anderson et al., 2002; Bodnar, Prahme, Cutting, Denckla, & Mahone, 2007; Toplak, West, & Stanovich, 2013; Vriezen & Pigott, 2002). One of the most commonly used executive function questionnaires for children is the Behaviour Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000), which also has a preschool version (BRIEF-P; Gioia, Espy, & Isquith, 2003).

Some researchers have reported that performance-based cognitive and executive function assessment results and BRIEF-P results are not strongly related (Pedersen, 2005; Rahbari & Vaillancourt, 2015). Other researchers, in contrast, have found that performance-based executive function tests relate to problem behaviours in real life, as reported by the parents of preschoolers on BRIEF-P questionnaires (Espy, Sheffield, Wiebe, Clark, & Moehr, 2011; Garon, Piccinin, & Smith, 2016). Thus, there is mixed evidence of how well performance-based executive function assessment results and questionnaires, such as the BRIEF-P, are related. One reason for this discrepancy is that studies investigating relationships between performance-based executive function assessments and questionnaires are still scarce in general, with even less known about their congruency in the preterm population. In the school-age preterm children, Ritter, Perrig, Steinlin, and Everts (2014) reported that performance-based assessment and questionnaires of executive functions did not significantly correlate. Similar results were noted by Loe and colleagues (2015): performance-based executive function measures were positively related to adaptive skills but not to parental reports of executive functions in three- to five-year-old preterm children. Furthermore, existing research has focused on correlations at a single point in time, offering limited insights into expected future executive function performance. The dearth of studies in this field, and the mixed results of those that do exist, make clinical assessments difficult for clinicians who are trying make decisions on which children are most at risk of executive dysfunction and in need of additional supports upon school entry. In Study 2, we researched how well performance-based executive function assessment results were related to real life executive function outcomes as reported by parents and teachers. This was to understand if executive function assessment at the preschool stage (four years) could predict future executive function difficulties when the children entered kindergarten at four to five years.

Parent and teacher reporting of executive functioning. Discrepancies in parent and teacher reporting of children's behaviour have been reported in the general population (Achenbach, McConaughy, & Howell, 1987). Parent and teacher reports of children's behavior were reported to greatly differ in a large multicultural study conducted by Rescorla et al. (2014), who found that parents tended to report more behavioural issues than teachers in school children over the age of six years. Other researchers have demonstrated that parents' and preschool child carers' reporting of behavioural difficulties differs even during the preschool years (Berg-Nielsen, Solheim, Belsky, & Wichstrom, 2012; Korsch & Petermann, 2014; Marković, Rescorla, Okanović, Maraš, Bukurov, & Sekulić, 2016). Overall, there seems to be fairly strong evidence that within the general population parents and teachers do not agree on children's behavioural presentation, and that this pattern also holds regarding the evaluation of children's executive functions. Some researchers, such as Pedersen (2005), have found that parents reported more executive function difficulties for young children than did teachers. In contrast, others have reported that parents identified fewer executive function problems than teachers (Mares, McLuckie, Schwartz, & Saini, 2007; Rahbari & Vaillancourt, 2015). Also, McCandless and O'Laughlin (2007) found a variable pattern of differences between parent and teacher reporting: parents reported more behavioural issues than teachers, but teachers reported more cognitive difficulties than parents. Overall, it is evident that teachers and parents tend to evaluate children's behaviour and executive function differently for general populations, but there are inconsistent findings regarding the number and types of issues parents and teachers report. Not surprisingly, externalizing problems, such impulsivity and aggression, tend to be easier to recognize and thus more uniformly reported than internalizing issues, such as anxiety and mental inflexibility (Kolko & Kazdin, 1993).

There is little knowledge relating to the parent and teacher reporting patterns of executive functions and behaviour of preterm children. The few studies that have been conducted in older school-age populations and young adults have mainly revealed that the parents of preterm subjects tend to report more executive function difficulties than parents of term subjects (Estroff, Yando, Burke, & Snyder, 1994; Heinonen et al. 2013). However, some researchers have argued that parents of preterm children do not report more difficulties than parents of term children (Schappin, Wijnroks, Uniken Venema, Jongmans, & Bruce, 2013).

The congruency between parent and teacher reporting of both executive functions and behaviour of four- to five-year-old preterm children has not been previously studied. As such, it is not clear whether findings from older preterm populations would hold for this group. It is important for clinicians working with preterm children to know if the parents and the teachers of preterm children report similar types of executive functions and behaviours, as caution would need to be applied in interpretation of the assessment results if there are reporting differences. Also, if there are differences in reporting, it is especially important to gain information from more than one source, otherwise the results of the assessment may be biased and not reflect the child's functioning across different settings. Thus, in the third paper, we looked at the congruence between parent and teacher reporting of executive function and behavioural difficulties of four- to five-year old preterm children.

Executive function theories and test selection. In our studies, we employed the theoretical frame work of Miyake et al. (2000) to cover the three components of EF: shifting, updating (working memory) and inhibition. The test selection within the study reflects the assessment tools used in clinical practice in order to enhance the applied nature of findings – in other words, to be relevant to current clinical practice. We wanted to understand if the performance-based assessment tools that were used in routine assessment were predictive of EF difficulties when the preterm children entered schooling. Firstly, we wanted to estimate the children's general intellectual functioning by using WPPSI-III subscales. The NEPSY-II narrative memory recall and sentence recall were selected as possible measures of working memory difficulties. The NEPSY-II word generation task can assess shifting in addition to language skills. The Shape School condition B measures inhibition, while condition C measures cognitive shifting. The Day-Night task is also used to measure shifting and inhibition abilities. Both Shape School conditions B and C and the Day-Night task require keeping rules in mind, placing demands also on working memory. In addition, EF and behavioural questionnaires were selected to cover both hot (behavioural and emotional) and cool (cognitive) aspects of EF.

Summary

Overall, while there is increasing evidence that young preterm children are vulnerable to developing executive function difficulties, it is unclear which medical and social risk factors are most likely to contribute to having executive dysfunction at the age of four to five years; the age at which children in some countries enter schooling. It is important to be aware of preterm children's difficulties prior to school, so possible intervention, educational supports, and extra funding can be put in place. Otherwise, the preterm children are at risk of falling behind their term peers academically and socially. Furthermore, little is known about the ecological validity of preschool executive function assessment tools in the context of performance-based vs. questionnaire assessments, and parent vs. teacher reporting of executive functions and behaviour. There is a need to consolidate which assessment tools could form the most valid battery to identify young preterm children's executive function and behavioural performance. Having internationally recognized guidelines relating to recommended assessment procedures could improve consistency of preterm children's assessments and inform the allocation of appropriate intervention resources. Also, there may be a need to develop better executive function and behaviour assessment tools for young children. The three studies in this thesis contribute to addressing the above issues. The preterm and term group characteristics, attendee and non-attendee preterm group characteristics and the sampling process are presented in the following Tables 1 and 2 and Figure 2.

Table 1 *Preterm and Control Group Characteristics*

	Preterm		Term		t
Boys, *n* (%)	70.00 (49.6)		45.00 (58.4)		1.55
Girls, *n* (%)	71.00 (50.4)		32.00 (41.6)		1.55
Age (months)at EF assessment (mean, range)	49.10 (48-58)		54.86 (48-67)		11.40***
Age (months) at parent questionnaires (mean, range)	58.33 (48-66)		57.35 (48-64)		-2.18
Age (months) at teacher questionnaires (mean, range)	58.40 (48-68)		58.41 (48-68)		-0.41
	M	*SD*	*M*	*SD*	χ^2
Social risk index (the below risks combined)	2.98	2.61	3.26	2.62	0.74
Maternal age	0.17	0.38	0.03	0.16	-3.94***
Family structure	0.36	0.75	0.42	0.71	0.54
Main carer education level	1.03	0.75	0.90	0.82	-1.20
Main income earner occupation	1.01	0.91	1.03	0.85	0.14
Main income earner work status	0.63	0.85	0.81	0.81	1.42
Language spoken at home	0.07	0.12	0.07	0.34	1.57

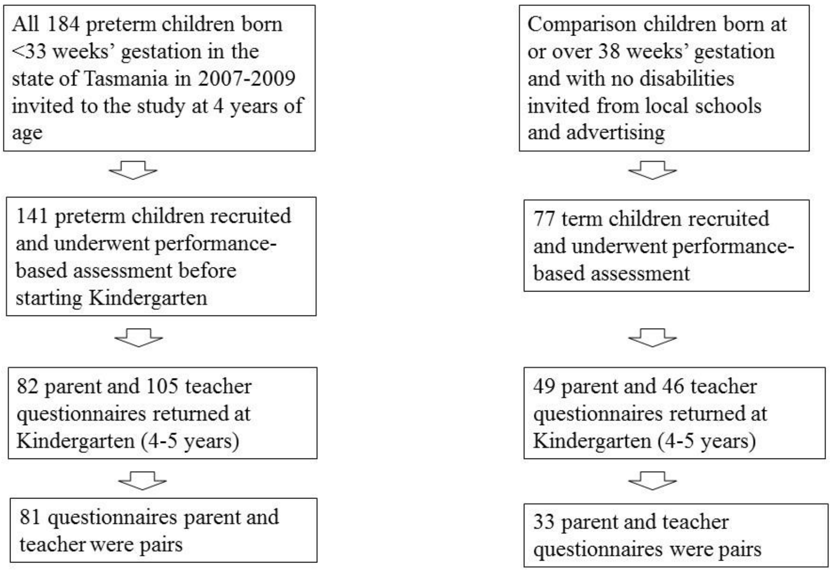
*p<.05, **p<.01, ***p<.001

Table 2 *Demographics and Neonatal Medical Conditions of Preterm Attendees (participants) and Preterm Non-attendees from the Initial Cohort of Preterm Children Born in 2007-2009*

	Preterm attendees (*n*=141)	Preterm non-attendees (*n*=43)	*p*
Boys, *n* (%)	70 (49.6)	25(58.1)	.332
Girls, *n* (%)	71 (50.4)	18 (41.9)	.332
Gestational age *M* (range)	29.40 (23-33)	30.37 (26-33)	.006**
Birthweight *M* (range)	1328.24 (530-2300)	1502.26 (700-2320)	.014*
Maternal age in years	27.79 (18-43)	27.77 (18-43)	.985
Sepsis *n* (%)	34 (24.1)	3 (7.0)	.002**
Necrotizing enterocolitis	8 (5.6)	0 (0)	.004**
Periventricular leukomalacia	20 (14.2)	4 (9.3)	.408
Intraventricular haemorrhage	5 (3.5)	0 (0)	.025*
Pulmonary dysplasia	19 (13.5)	1 (2.3)	.003**
Other brain condition	7 (4.9)	0 (0)	.008**
Post-natal steroids	3 (2.1)	0 (0)	.083
Intrauterine growth restriction	10 (7.1)	4 (9.3)	.659

*p<.05, **p<.01, ***p<.001

Figure 2. **Sampling process**



Chapter 2. Risk factors for executive function difficulties in preschool and early school-age preterm children

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Risk factors for executive function difficulties in preschool and early school-age preterm children

Abstract

Aim*:* To investigate the relationship between executive functioning and social and perinatal risk factors in four- to five-year-old preterm children.

Methods*:* 141 children born preterm (< 33 weeks of gestation) and 77 term comparison children were assessed using standardized measures of general intelligence and performance-based executive function tests prior to starting kindergarten. Parent and teacher reports of executive functioning were completed when the children commenced kindergarten. The preterm and the term comparison groups were compared on measures of intelligence and executive functions using independent groups *t*-tests, and multivariate regression analyses were performed to identify factors predictive of intelligence and executive functioning in the preterm group.

Results: The preterm group performed significantly more poorly than the comparison group on all intelligence and executive function tests. The parental reports of the preterm and term comparison children's executive function did not differ significantly, but the teachers reported elevated executive function difficulties for the preterm group. Higher social risk, in particular lower educational level of the main caregiver, was the strongest predictor for the preterm children's intelligence and executive function results.

Conclusion*:* Social risk factors are strongly associated with impaired early executive function outcomes in preterm children.

Background

Preterm birth is defined as birth occurring before 37 weeks' gestation (1). It is known that school-age children born preterm are at increased risk of cognitive problems compared to their full-term peers, including executive function (EF) difficulties (2, 3). Executive functions, such as working memory, self-control, cognitive flexibility and organisational skills, form an important basis of successful entry to school. In fact, it has been reported that executive functions are more strongly associated with school readiness than is general intelligence (IQ) (4). Nevertheless, many preterm children are not routinely monitored in terms of their development, and when it does occur, surveillance is often limited to the first couple of years of preterm children's lives. This is largely due to lack of resources, but also a limited understanding of the persistence of higher order cognitive difficulties, which can occur despite acceptable developmental progress in infancy.

Given the importance of executive functions for the transition to school in preterm children, the early detection of children at high-risk of EF difficulties has implications for surveillance and early intervention. There are numerous early medical and demographic factors that may help identify high-risk children, including gestational age, male sex, higher social risk, and neonatal complications associated with longer hospital stays (e.g., brain injury, and infections). While previous research has demonstrated that these factors predict school-age cognitive outcomes (5-8), it is less clear whether they are predictive of preschool general intelligence and executive functioning. Some researchers have found associations between medical/demographic factors and EF in preschool populations (e.g., 9,10), but these studies have often used less well-known tasks, rather than standardized assessment tools. It is important to gain a better understanding how we can utilize both the knowledge of medical and demographic risk factors and clinical assessment results to identify children most at risk. Early identification of children at risk of EF and cognitive deficits allows for intervention and remediation prior to school-entry, thereby potentially reducing adverse effects on educational and academic attainment.

The main aim of the current study was to examine the association of social and perinatal risk factors with cognitive functioning in preschoolers born preterm, with a focus on executive functioning. We measured a range of executive function components, and utilized both performance-based and questionnaire outcomes. On the basis of previous findings with school-aged children, we hypothesised that earlier gestational age, higher social risk, male sex, and longer hospital stay would be predictive of lower IQ and poorer executive functioning. An additional aim was to assess the magnitude of the cognitive deficits in the preterm group by comparing them to a term comparison group.

Participants and assessment process

Preterm children eligible for this study were born at less than 33 weeks' gestation and cared for at the Royal Hobart Hospital (RHH) (*n*=184) in 2007-2009. After University ethics committee approval, 141 children (77%) were recruited from the routine follow-up of preterm infants offered by the RHH Neonatal Intensive Care Unit Follow-up Clinic as close as possible to their fourth birthday. As this study had a strongly clinical focus, age was not corrected for prematurity. In clinical practice age is corrected for prematurity up to two to three years but not beyond (11), and generally there is no extra consideration for degree of prematurity in the education system. Ten children were not contactable or had moved away, six declined to participate in the study, and 27 did not attend after multiple reminders. The mean gestational age of the participating preterm children was 29.69 weeks (range 23.6-32.5 weeks), and none had congenital syndromes. Children who could not participate in subtests due to significant global delay or sensorimotor issues (*n*=5) were given the minimum score on those subtests.

The four- to five-year-old comparison group participants (*N*=77) were recruited from local schools (pre-kindergarten groups and kindergarten, i.e., prior to compulsory formal schooling), and by advertising at the RHH. All were born at or over 38 weeks' gestation and had no diagnosed disabilities. The preterm and comparison groups were matched for the age at the time of the questionnaire completion, sex distribution and social risk. However, the preterm group was younger at the time of the performance-based assessment. This was partly mitigated by using age-standardized scores or controlling for age. Table 1 shows the preterm group and comparison group characteristics.

The children's socioeconomic risk was determined by a social risk index (12). The six risk factors are maternal age at the time of birth, family structure, main carer education level, main income earner occupation, main income earner employment status, and language spoken at home, all of which have a risk scale from 0 (low risk) to 2 (high risk). The total score was calculated by combining the six factors. We used the length of hospital stay as an overall indicator of medical risk. There were insufficient numbers in this study to compare separate medical complications such as brain injury or infections.

Performance-based assessment (four years)

At four years, the preterm children underwent performance-based intellectual and executive function assessment. General intelligence was assessed with the *Wechsler Preschool and Primary Scale of Intelligence, 3rd edition* (WPPSI-III) (13), and the cognitive functioning of the preterm and comparison group was compared on the subtests of Block Design, Matrix Reasoning, Information and Coding. Due to logistical issues, a small number of term comparison children (*n*=9) were older than four years when completing the performance-based assessments.

To assess executive functioning, subtests from the *Developmental Neuropsychological Assessment battery* (NEPSY-II; 14) were administered to both groups (narrative memory recall, sentence recall and word generation), along with the *Shape School Task* (15), and the *Day-Night Stroop* (16). The *Shape School Task* is a measure of inhibition, switching set and combination of both skills. It is a storybook-like assessment tool for preschoolers with human-like coloured shape figures. In condition A (control measure for baseline naming speed), the child is told that the figure's name is the colour, and the child has to say the name (colour) as quickly as possible without making any errors. In Condition B (Switch), the figures have both happy and sad faces. The child is told to name only the shape of the figures that are happy and inhibit saying the names of the sad shapes. In conditions C and D, some figures are wearing hats. In condition C, the child has to say the colour of the figures with hats and the shape for figures without hats, measuring cognitive shifting. Both conditions B and C require keeping two rules in mind, placing demands on working memory. The *Day-Night Stroop* can be used with young children to measure switching and inhibition abilities. In this test, the child is required to say "day" when presented a page showing a night-time sky and "night" when shown a picture of a sun (16 trials). The WPPSI-III and NEPSY-II provide standardized age norms. For the Day-Night and Shape School tasks we used raw scores, but age-controlled scores when comparing the preterm and term groups.

Questionnaire assessment (four to five years)

The year following the performance-based cognitive assessment, when the preterm children were four to five years old and had started kindergarten, their parents and teachers were sent the *Behavior Rating Inventory of Executive Function – Preschool Version* questionnaires (BRIEF-P; 17). This is a rating scale developed to measure everyday behaviours associated with specific areas of executive functioning in children aged two to five years. It has five subscales: Inhibit, Shift, Emotional Control, Working Memory and Plan/Organize. The scales have three summary indexes: the Inhibitory Self-Control Index, the Flexibility Index, and the Emergent Metacognition Index. Age-standardized scores were used in the study. The questionnaires were not provided at the time of the performance-based assessments as the children were not yet at school and we wanted concurrent reports from parents and teachers. Parents and teachers of both groups were informed that the study's aim was to compare the higher cognitive functioning of preterm and term children. While the teachers were not specifically informed if the child was born preterm, they may have had that knowledge. The teacher questionnaires were completed three to five months after the start of the school year. We had high return rates: 95% of parent and 75% of teacher questionnaires.

Results

Between-group comparisons (preterm vs. comparison) on standardized measures of IQ and EF were assessed on all measures using independent-groups *t*-tests, with Bonferroni correction for multiple comparisons and controlling for the effects of age where necessary. The preterm group performed significantly more poorly than the comparison group on all intelligence and executive function tests (effect size g=0.49 to 1.5; see Table 2). As standardized scores are not available for the Day-Night and Shape School tasks, analyses of covariance were performed, controlling for age. All statistical differences persisted (Table 2). Based on parental report, there were no significant group differences on the BRIEF-P (g=0.00 to 0.24). However, the teachers reported elevated difficulties for the preterm group on several subscales: inhibition, working memory, planning/organizational skills, self-control and overall emergent metacognitive skills (g=0.42 to 0.64; see Table 3).

Simultaneous multiple regression analysis was used to examine factors that may predict IQ and EF in the preterm group (gestational age, birthweight, social risk, sex, and length of hospital stay). Due to issues of multicollinearity, we assessed the effects of gestational age and length of stay separately, using two regression models. Gestational age is a well-known predictor for outcomes of preterm children, but length of stay in hospital had the highest number of correlations with the IQ and EF outcomes in our study. Thus, we first investigated the associations of gestational age, sex and overall social risk level with the cognitive and executive function outcome measures by using simultaneous regression analysis. Next, we used the length of hospital stay instead of gestational age. Higher social risk was independently associated with all intellectual measures (standardized β=-0.22 to -0.52), performance-based executive function assessment results (standardized β=-0.24 to -0.42), and most parent and teacher questionnaire results in both models (standardized β=0.30 to 0.51). Male sex independently predicted poorer outcome on four mainly verbal subscales on the WPPSI-III, lower verbal and full scale IQ and memory (standardized β=0.16 to 0.32). Gestational age was independently associated with only five out of 18 performance-based measures (mainly those including naming and processing speed; standardized β=0.16 to 0.25), and it had some association with the questionnaire scores (standardized β=-0.24 to 0.38). Length of hospital stay was independently associated with some IQ measures (standardized β=-0.17 to -0.30), two performance-based EF measures (standardized β=-0.24 to -0.25), and some reported EF difficulties (standardized β=0.22 to 0.37). To investigate social risk in more detail, we conducted a further multiple regression analysis in which we entered all the separate social risk factors simultaneously. The analysis indicated that the main carer education level was the strongest predictor for executive function (standardized β=-0.22 to 0.40).

Discussion

The current study aimed to identify predictors of executive function difficulties in school-entry preterm children. The preterm group performed significantly more poorly than the term comparison group on direct measures of IQ and EF. These results are similar to those reported in older preterm children (2, 18). This robust finding supports our premise that cognitive difficulties in preterm children are evident at preschool age, and emphasises the need to develop preventive and remedial measures to reduce the discrepancy between preterm and full-term children. However, there were conflicting results between parents and teachers who rated executive functioning in everyday settings. Specifically, the parents in the preterm group and the parents in the comparison group reported their children as having similar rates of difficulty. In contrast, the teachers of the preterm children reported the children as having more difficulties with inhibition, working memory, planning/organizational skills and self-control than did the teachers of the comparison group. A possible explanation for this discrepancy is that parents of the preterm children may feel encouraged by the early developmental outcomes of their children, who may have been given initially a cautious or negative prognosis, and thus they may overestimate the higher-level cognitive abilities of their children. Alternatively, executive function difficulties may not be evident in the preterm four- to five-year-olds at home, but they may be more evident in the school setting in which children are required to be more focussed and organized and to comply with group rules and instructions. The teachers may be more perceptive of mild cognitive and behavioural difficulties than parents, given teachers' more extensive experience.

Contrary to our expectations, only social risk was strongly associated with all executive functions. Low educational level of the main caregiver (i.e. less than 11 years of education) was the strongest social predictor of poorer executive function. This finding is in agreement with general population studies of children from higher social risk backgrounds having less proficient executive functions (6, 19), and with reports that socio-economic environment has a greater impact on cognitive outcomes than the genetic profile of the child (20). There is some previous evidence of lower parental education level impacting negatively on the development of children's executive functions (21, 22), with the current study contributing in this regard. However, more studies are needed, especially in the preterm preschoolers, to investigate the association. Also, we did not assess the intellectual and executive functioning of the caregivers of the children in the current study. Such further research could clarify how much impact the genetically inherited intellectual capacity and the caregivers' executive function skills may have on the development of preterm children's executive functions compared to other social, medical and educational factors. We did not separately control the preterm children's IQ as IQ overlaps with EF, and using it as a co-variate can produce overcorrected findings about neurocognitive function (23).

Unexpectedly, gestational age and the length of hospital stay were not independent predictors of executive function difficulties in our preterm group. Many studies have shown that the risk of developmental and cognitive difficulties increases with decreasing gestational age (5, 24). Nevertheless, some researchers have not found gestational age to be such a strong predictor (25, 26). While gestational age has been shown to have a clear association with survival rates and severe neurodevelopmental delays of preterm children, it is possible there is more variability in how the gestational age impacts on higher cognitive processes, especially in younger children. Commonly recognized risk factors like gestational age, medical complications, and sex of a child cannot be modified. However, our findings offer a more positive message for parents of preterm children and professional working with them: there are other factors, such as social risk, that can be possibly be mitigated, providing better outcomes for preterm children.

The question remains as to whether children born in recent decades have a different outcome to the children born in the previous century, due to improvements in medical and therapeutic care. It should be noted that all preterm children in our study had access to regular medical/allied health surveillance and free preschool educational/allied health therapies provided by the state. Further studies on the effect of preterm child follow-up and associated interventions could strengthen understanding of its relevance to EF outcomes.

In summary, these results emphasize the importance of the social environment on the development of preterm preschoolers' executive functions. There is a need for research examining why the children from families at greater social risk have poorer outcomes, to enable the establishment of possible interventions to assist these vulnerable children. There are psychosocial, parenting and educational intervention programmes that have been shown to have a positive impact on cognitive and behavioural development, and to improve executive functioning of children living in high social risk families in general populations (27, 28). Nevertheless, intervention programs aiming to improve the cognitive and executive function outcomes of preterm children have not generally been proven to have long-term effects (29, 30). There is a clear need for more effective identification of higher-level cognitive difficulties prior to preterm children entering school, especially when they come from families with high social risk. Such identification would allow for intervention, remediation and support prior to school-entry, thereby reducing potential effects on educational and academic attainment.

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Conflict of interest statement

There are no conflicts of interest

Ethics committee approval

This study was approved by the Tasmanian Human Research Ethics Committee (H0011567) and the Tasmanian Social Science Ethics Committee (H0014174).

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Table 1 *Preterm and Control Group Characteristics*

	Preterm		Comparison		*t*
Boys, *n* (%)	70.00 (49.6)		45.00 (58.4)		1.55
Girls, *n* (%)	71.00 (50.4)		32.00 (41.6)		1.55
Age (months)at EF assessment (mean, range)	49.10 (48-58)		54.86 (48-67)		11.40***
Age (months) at parent questionnaires (mean, range)	58.33 (48-66)		57.35 (48-64)		-2.18
Age (months) at teacher questionnaires (mean, range)	58.40 (48-68)		58.41 (48-68)		-0.41
	M	*SD*	*M*	*SD*	χ^2
Social risk index (the below risks combined)	2.98	2.61	3.26	2.62	0.74
Maternal age	0.17	0.38	0.03	0.16	-3.94***
Family structure	0.36	0.75	0.42	0.71	0.54
Main carer education level	1.03	0.75	0.90	0.82	-1.20
Main income earner occupation	1.01	0.91	1.03	0.85	0.14
Main income earner work status	0.63	0.85	0.81	0.81	1.42
Language spoken at home	0.07	0.12	0.07	0.34	1.57

*p<.05, **p<.01, ***p<.001

Table 2 Intelligence and executive function assessment

	Preterm (*n*=141)		Comparison (*n*=77)					
	M	SD	M	SD	*t/F**	*p*	*df*	*g*
<u>*WPPSI-III*</u>								
Block Design[a]	8.11	3.17	10.55	3.10	4.82	<.001	184	0.77
Information[a]	9.05	4.10	11.05	2.64	3.96	<.001	152	0.54
Matrix Reas[a]	8.47	3.17	10.46	2.91	4.03	<.001	184	0.64
Coding[a]	7.05	3.55	10.95	2.37	8.72	<.001	149	1.20
<u>*NEPSY-II*</u>								
Narrative Recall[a]	5.23	2.33	8.77	3.25	8.17	<.001	170	1.35
Sentence Recall[a]	7.21	3.86	11.25	2.90	7.40	<.001	129	1.12
Word Gen[a]	8.04	3.38	12.07	2.34	9.08	<.001	140	1.29
Day-Night Efficiency[b]	0.18	0.12	0.37	0.15	8.60	<.001	136	1.50
(adjusted by age)	0.18	0.02	0.37	0.02	37.2	<.001	136	0.95
Shape A Efficiency[b]	0.55	0.24	0.78	0.28	5.30	<.001	145	0.49
(adjusted by age)	0.58	0.03	0.73	0.04	6.15	.014	145	0.44
Shape B Efficiency[b]	0.44	0.22	0.66	0.30	4.73	<.001	135	0.88
(adjusted by age)	0.46	0.03	0.63	0.04	8.67	.004	135	0.50
Shape C Efficiency[b]	0.18	0.09	0.28	0.12	5.00	<.001	116	0.92
(adjusted by age)	0.19	0.01	0.28	0.02	12.2	.001	116	0.59

**F*-values provided for age-adjusted scores, analysed by ANCOVA

Table 3 Parent and teacher reporting of executive functioning (BRIEF-P, scaled scores)

	Preterm		Comparison					
	M	SD	M	SD	*t*	*p*	*df*	*g*
Parents	(*n*=81)		(*n*=49)					
Inhibit	52.50	12.06	51.00	11.37	-0.71	.478	128	0.13
Shift	49.63	11.15	47.98	8.62	-0.89	.376	128	0.16
Emotional Control	50.90	12.69	50.65	11.69	-0.11	.912	128	0.02
Working Memory	55.48	15.61	52.04	11.68	-1.33	.155	122	0.24
Plan/Organize	51.62	13.97	51.61	13.12	-0.00	.998	128	0.00
Self-Control	51.88	13.18	50.04	11.07	-0.82	.416	128	0.15
Flexibility	49.99	12.62	49.06	10.69	-0.43	.669	128	0.08
Emergent Metacognitive	54.23	15.64	51.61	12.38	-1.06	.293	119	0.18
Teachers	(*n*=105)		(*n*=46)					
Inhibit	50.92	11.89	45.43	8.80	-2.81	.006	149	0.50
Shift	47.48	9.11	45.83	7.01	-1.09	.276	149	0.19
Emotional Control	46.15	7.31	45.46	7.39	-0.54	.592	149	0.09
Working Memory	54.09	12.34	47.00	7.57	-4.32	<.001	132	0.64
Plan/Organize	52.92	14.13	45.20	8.76	-4.09	<.001	132	0.60
Self-Control	48.86	10.14	44.65	9.49	-2.39	.018	149	0.42
Flexibility	46.92	8.57	44.83	7.35	-1.44	.151	149	0.25
Emergent Metacognitive	53.87	13.50	46.11	7.95	-4.40	<.001	136	0.64

Appendix S1 Predictors for intellectual and executive functioning of preterm children
Regression model 1 (gestational age, social risk and sex)

		b	Lower limit 95%CI	Upper limit	β
Social risk					
WPPSI-III	Information	-0.81	-1.04	-0.58	-
	Verbal IQ	-6.02	-7.87	-4.17	-
	Word Reasoning	-0.66	-0.86	-0.45	-
	Vocabulary	-0.60	-0.80	-0.39	-
	Matrix	-0.49	-0.68	-0.30	-
	Full Scale IQ	-4.37	-6.20	-2.52	-
	Performance IQ	-3.40	-4.98	-1.81	-
	Picture Concepts	-0.36	-0.54	-0.18	-
	Coding	-0.41	-0.63	-0.20	-
	Block Design	-0.36	-0.56	-0.16	-
NEPSY-II	Word	-0.55	-0.76	-0.33	-
	Sentence Recall	-0.61	-0.86	-0.36	-
	Narrative Recall	-0.29	-0.45	-0.13	-
	Narrative Recog	1.18	0.39	1.90	-0.25*
Shape School	Naming	-0.05	-0.07	-0.03	-
	Inhibition	-0.04	-0.06	-0.03	-
	Switching	-0.02	-0.02	-0.01	-
Day-Night task	Efficiency	-0.02	-0.03	-0.01	-
BRIEF-P parent	Planning	0.93	0.59	1.26	0.51**
	Inhibition	1.38	0.85	1.90	0.49**
	Emotional	0.81	0.48	1.13	0.48**
	Shift	0.69	0.36	1.03	0.41**
	Working	1.22	0.62	1.82	0.39**
BRIEF-P teacher	Planning	0.72	0.37	1.06	0.38**
	Working	1.11	0.58	1.65	0.38**
	Inhibition	0.76	0.23	1.28	0.27**
Gestational age					
WPPSI-III	Picture Concepts	0.31	0.11	0.51	0.24***
	Coding	0.34	0.11	0.58	0.23**
	Vocabulary	0.24	0.01	0.47	0.16*
Shape School	Naming	0.03	0.01	0.05	0.25**
NEPSY-II	Word Generation	0.31	0.09	0.54	0.22**
BRIEF-P parent	Working	-1.02	-1.69	-0.35	-0.30*
	Shift	-0.51	-0.89	-0.14	-0.27**
	Inhibition	-0.78	-1.36	-0.20	-0.25**
	Emotional	-0.44	-0.80	-0.08	-0.24*
BRIEF-P teacher	Working	1.11	0.58	10.65	0.38**
	Planning	0.72	0.37	1.06	0.38**
Sex					
WPPSI-III	Vocabulary	1.80	0.73	2.87	0.25**
	Word Reasoning	1.71	0.64	2.77	0.24**
	Verbal IQ	15.25	5.70	24.80	0.23**
	Coding	1.62	0.51	2.73	0.23**
	Full Scale IQ	12.11	2.54	21.68	0.20**
	Information	1.53	0.36	2.70	0.19*
NEPSY-II	Narrative Recog	2.27	1.00	3.54	0.30**
	Narrative Recall	1.18	0.39	1.90	0.25*

	Sentence Recall	1.67	0.37	2.97	0.21*
Day-Night task	Efficiency	0.04	0.00	0.08	0.16*

*p<.05, **p<.01, ***p<.00

Appendix S2 Predictors for intellectual and executive functioning of preterm children
Regression model 2 (length of stay in hospital, social risk and sex)

		b	Lower limit 95%CI	Upper limit	β
Social risk					
WPPSI-III	Information	-0.73	-0.98	-0.50	-
	Verbal IQ	-5.61	-7.61	-3.62	-
	Word Reasoning	-0.62	-0.84	-0.40	-
	Vocabulary	-0.57	-0.80	-0.35	-
	Matrix	-0.40	-0.60	-0.20	-
	Full Scale IQ	-3.61	-5.60	-1.62	-
	Performance IQ	-2.53	-4.17	-0.89	-0.27**
	Picture Concepts	-0.28	-0.48	-0.09	-0.25**
	Coding	-0.30	-0.53	-0.08	-0.22**
	Block Design	-0.25	-0.46	-0.05	-0.22*
NEPSY-II	Word	-0.50	-0.73	-0.27	-
	Sentence Recall	-0.54	-0.82	-0.27	-
	Narrative Recall	-0.27	-0.45	-0.10	-0.28**
	Narrative Recog	-0.38	0.65	-0.11	-0.24**
Shape School	Inhibition	-0.04	-0.06	-0.02	-
	Naming	-0.04	-0.06	-0.02	-
	Switching	-0.01	-0.02	-0.01	-
Day-Night task	Efficiency	-0.02	-0.02	-0.01	-
BRIEF-P parent	Inhibition	1.40	0.85	1.95	0.50**
	Planning	0.87	0.54	1.20	0.48**
	Emotional	0.76	0.43	1.09	0.46**
	Shift	0.68	0.32	1.03	0.41**
	Working	1.12	0.50	1.74	0.36**
BRIEF-P teacher	Planning	0.69	0.32	1.07	0.37**
	Working	1.03	0.47	1.60	0.35**
	Inhibition	0.84	0.27	1.415	0.30**
Length of stay					
WPPSI-III	Picture Concepts	-0.02	-0.03	-0.01	-0.30***
	Block Design	-0.20	-0.03	-0.01	-0.27**
	Coding	-0.02	0.04	-0.01	-0.25**
	Performance IQ	-0.12	-0.22	-0.01	-0.20*
	Information	-0.02	-0.03	-0.00	-0.17*
NEPSY-II	Word Generation	0.02	-0.04	-0.01	-0.24**
Shape School	Naming	-0.00	-0.00	-0.00	-0.25**
BRIEF-P parent	Working	0.08	0.04	0.13	0.37*
	Planning	0.05	0.02	0.70	0.36**
	Emotional	0.04	0.01	0.06	0.30**
	Inhibition	0.50	0.01	0.10	0.26**
BRIEF-P teacher	Working	0.50	0.01	0.10	0.22*
Sex					
WPPSI-III	Vocabulary	1.84	0.74	3.01	0.26**
	Verbal IQ	16.99	6.88	27.06	0.26**
	Word Reasoning	1.85	0.72	2.98	0.25**
	Coding	1.70	0.53	2.85	0.24**
	Information	1.83	0.62	3.04	0.22**

	Full Scale IQ	12.88	2.80	22.95	0.21*
NEPSY-II	Narrative Recog	2.50	1.13	3.86	0.32**
	Narrative Recall	1.21	0.35	2.07	0.25**
	Sentence Recall	1.66	0.26	3.06	0.21*
Day-Night task	Efficiency	0.04	0.00	0.08	0.17*

*p<.05, **p<.01, ***p<.001

Appendix S3 Most significant predictors for intellectual and executive functioning of preterm children

Regression model 3 (separate social risk factors)

		b	Lower limit 95%CI	Upper limit	β
Main carer education					
WPPSI-III	Full Scale IQ	-11.34	-19.66	-3.03	-0.26**
	Verbal IQ	-12.23	-20.72	-3.75	-0.26**
	Performance IQ	-9.32	-16.53	-2.11	-0.25*
	Matrix	-1.11	-1.95	-0.28	-0.24**
	Information	-1.27	-2.30	-0.24	-0.22*
	Word Reasoning	-1.14	-2.08	-0.19	-0.22*
	Coding	-1.17	-2.09	-0.14	-0.22*
	Vocabulary	-1.03	-2.02	-0.04	-0.20*
Day-Night	Efficiency	-0.05	-0.10	-0.01	-0.31*
NEPSY-II	Word	-1.06	-2.03	-0.09	-0.22*
BRIEF-P	Emotional	2.86	1.22	4.50	0.40
	Flexibility	4.95	1.66	8.23	0.36**
	Shift	2.14	0.41	3.87	0.30*
	Planning	1.90	0.14	3.66	0.25*
	Working	3.14	0.02	6.25	0.24*
	Self-Control	4.25	0.44	8.06	0.24*
BRIEF-P	Flexibility	3.28	1.25	5.30	0.35**
	Shift	1.57	0.45	2.69	0.32**
	Working	3.78	1.34	6.22	0.32**
	Planning	2.23	0.65	3.80	0.30**
	Emotional	1.32	0.25	2.40	0.28*
	Metacognitive	5.27	1.29	9.25	0.28**
	Inhibition	2.97	0.57	5.37	0.27*
	Self-Control	3.84	0.58	7.10	0.27*
Empl status					
WPPSI-III	Information	-1.61	-2.57	-0.66	-0.34**
	Vocabulary	-1.10	-2.03	-0.02	-0.26*
	Word reasoning	-1.08	-1.96	-0.19	-0.25*
	Coding	-1.06	-1.97	-0.14	-0.25*
	Matrix	-0.88	-1.66	-0.10	-0.24*
	Verbal IQ	-9.37	-17.28	-1.46	-0.24*
BRIEF-P	Shift	3.59	1.05	6.13	0.40**
	Emotional	1.83	0.21	3.44	0.34 *
	Flexibility	3.48	0.21	6.75	0.33*
	Self-Control	4.26	0.49	8.02	0.32*
Language					
WPPSI-III	Matrix reasoning	-6.02	-9.89	-2.15	-0.23**
	Information	-5.60	-10.33	-0.80	-0.17*
BRIEF-P	Emotional	9.21	3.18	15.24	0.31**
	Planning	13.50	4.56	22.41	0.28**
	Self-Control	23.69	5.45	41.94	0.25*
	Working	17.36	3.20	31.51	0.22*
	Shift	6.68	0.25	13.12	0.21*
	Inhibition	14.50	0.50	28.50	0.20*

**p*<.05, ***p*<.01, ****p*<.001

Chapter 3. Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice.

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Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice

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Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice

Abstract: The aim of this study was to determine whether specific performance-based executive function assessment tools were associated with executive functioning in everyday life as reported by parents and teachers of four- to five-year-old preterm and term children. At the age of four years, 141 preterm children born <33 weeks' gestation and 77 term children were assessed using performance-based intelligence (WPPSI-III) and executive function (EF) assessment tools (NEPSY-II, Day-Night and Shape School tasks). The assessment results were compared to the parent and teacher completed questionnaires of EF (BRIEF-P) when the children started kindergarten at the age of four to five years. The performance-based EF assessment results were not consistently associated with the parent and teacher reports of EF in everyday life for either preterm or term groups. Clinical implications of using and interpreting performance-based EF assessment tools and EF questionnaires are discussed with a particular focus on young preterm children at the commencement of formal schooling.

Keywords: executive functions; preterm; questionnaire; assessment; children

Introduction

Executive functions (EF) are a set of higher cognitive skills including mental set shifting, working memory and inhibition (Huizinga, Dolan, & van der Molen, 2006), which are needed for purposeful, goal-directed behavior (Lezak, 1983). Despite growing scientific interest in the EF of preschool and early school age children, it is still uncommon to assess EFs in children under the age of six years in clinical practice, even in high-risk groups, such as those born preterm. This is due, in part, to the scarcity of suitable tools to assess young children's EFs (Anderson & Reidy, 2012). Furthermore, the ecological validity of EF assessment tools, that is, how well assessment results relate to and predict real-life functioning (Sbordone & Long, 1998), has not been widely evaluated in this age group. Ecologically valid findings are important for identification of young children at risk of EF difficulties, as such difficulties can interfere with functioning in everyday life. In this article, we investigate the associations between the performance-based EF assessment results and the parental and teacher reports of EF in everyday life for four- to five-year-old preterm and term groups.

The early detection of EF difficulties is especially important for children whose brain development has potentially been interrupted, for example by preterm birth. Preterm birth refers to a birth occurring before 37 weeks' gestation (Goldenberg, Culhane, Iams, & Romero, 2008). It is known that children sustaining brain insults prior to and around the time of birth, such as those associated with preterm birth, are most at risk for EF problems (Anderson et al., 2010; Mulder, Pitchford, Hagger, & Marlow, 2009). Being born early interferes with programmed brain development processes including the differentiation of pre-myelinating oligodendrocytes, growth of microglia, axons, and subplate neurons, expansion of the thalamic nuclei, and increase in cortical surface area and gyral formation in the cerebral cortex (Nosarti et al., 2011). Additionally, preterm children are at high risk of having perinatal brain injuries, such as intraventricular haemorrhages, periventricular leukomalacia and diffuse white matter abnormalities (Kapelloue et al., 2006; Volpe, 2001, 2009). Subsequently, preterm children's odds of having cognitive difficulties, including altered EF development, are high.

While 'executive functions' is used as an umbrella term, a further distinction is also made between cognitive ('cool') and social-emotional ('hot') executive functions (Hongwanishkul, Happaney, Lee, & Zelazo, 2005). 'Cool' executive functions refer to cognitive processes, such as attention, working memory, planning and monitoring performance. 'Hot' executive functions refer to social and emotional domains, such as mood, emotional control and social information processing (Hongwanishkul et al., 2005). While there are no purely 'hot' or 'cool' EF tasks, there is growing evidence that 'hot' and 'cool' processes are correlated but have distinct functions (Brock, Rimm-Kaufman, Nathanson, &

Grimm, 2009; Willoughby, Kupersmidt, Voegler-Lee, & Bryant, 2011). Comprehensive evaluation of EFs in young preterm children is especially important, as they are at an increased risk of both 'hot' and 'cool' EF difficulties when compared to term children (Anderson & Doyle, 2004; Deobel-Ayoub et al., 2009; Samara, Marlow, & Wolke, 2008). Nevertheless, it remains unclear which assessment tools best capture all aspects of EF in young children and how well currently available psychometric assessment tools predict reported everyday functioning, especially in the preterm population.

Furthermore, many preterm children are not routinely followed up, and for those that are monitored the follow-up is most likely restricted to the first few years of life. If preterm children have met their basic developmental milestones, such as walking and talking, they are usually discharged from follow-up without consideration of possible higher level cognitive difficulties which may emerge later (Salt & Redshaw, 2006). Follow-up assessments generally entail developmental or intelligence tests, but these tests correlate weakly to moderately with specific EF measures (dos Santos, de Kieviet, Königs, van Elburg, & Oosterlaan, 2013; Friedman et al., 2006). Thus, it can be difficult to reliably predict the outcome of preterm children's executive functioning when they enter schooling based solely on their early developmental or basic intelligence assessments. However, due to the limited availability of commercially available EF assessment tools for preschoolers, and possibly also lack of awareness of the importance of assessing EFs in young children, psychologists and even neuropsychologists may not evaluate preterm children's EFs prior to starting school. Opportunities to formulate recommendations relating to possible funding, interventions and supports when transitioning to school life may thus be missed.

The most common ways to assess children's EFs are performance-based testing, parent and teacher interviews, and standardized rating scales completed by parents and/or teachers (Mahone & Schneider, 2012). While studies have reported EF deficits in cohorts of preterm children (Taylor, Klein, Drotar, Schluchter, & Hack, 2006), very few have focussed on the level of agreement between performance-based measures of EF and functional difficulties in self-control, working memory, planning and organisation in the preterm population (Scott et al., 2012). Also, there is little knowledge about which cognitive assessment measures prior to commencement of schooling predict everyday EF difficulties in the classroom. It is important to know how well a child's performance-based assessment results correlate with everyday functioning, so that reliable and valid information can be provided to parents, teachers and other people working with the child. Furthermore, psychologists need to apply caution when interpreting the performance-based assessment results if they do not predict everyday functioning. Overestimating children's EF difficulties may cause unnecessary concern for parents and waste valuable therapeutic and educational resources. More importantly, children needing such supports may not be correctly identified if assessment tools underestimate their difficulties.

There has been limited research on the ecological validity of psychometric assessment tools used across the lifespan. The few published adult studies have reported poor correspondence between performance-based assessment tools and everyday functioning as measured by rating scales (Chaytor & Schmitter-Edgecombe, 2003; Higginson, Arnett, & Voss, 2000; Spooner & Pachana, 2006). There are even fewer studies regarding the ecological validity of paediatric psychometric assessment tools, especially relating to preschoolers and preterm children (Olson, Jacobson, & Van Oot, 2013). One of the rare reviews related to ecological validity of neuropsychological assessment measures that included paediatric data found that ecologically valid assessment tools for use with children are scarce (Rabin, Burton, & Barr, 2007). Also, the validity of EF questionnaires has also been questioned. A commonly used EF questionnaire is the Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000), which has a preschool version (BRIEF-P; Gioia, Espy, & Isquith, 2003). While studies have reported the BRIEF to correlate with performance-based EF (Espy, Sheffield, Wiebe, Clark, & Moehr, 2011; Garon, Piccinin, & Smith, 2016; Rabin, Burton, & Barr, 2007), this is not a universal finding (Conklin, Salorio, & Slomine, 2008; McAuley, Chen, Goos, Schachar, & Crosbie, 2010; Pedersen, 2010; Sølsnes et al., 2014; Toplak, West, & Stanovich, 2013). Mixed results have been reported in a wide range of studies looking at the associations between BRIEF and performance-based assessment in other paediatric populations, such as children with traumatic brain injury (Anderson, Anderson, Northam, Jacobs, & Mikiewicz, 2002) and neurofibromatosis (Payne, Hyman, Shores, & North, 2011). Furthermore, the underlying constructs of the BRIEF have been questioned (Bodnar, Prahme, Cutting, Denckla, & Mahone, 2007; Fuhs & Day, 2011; Spiegel, Lonigan, & Phillips, 2017). Overall, it appears that EF questionnaires can provide some indication of a person's EF capacity, as evidenced by associations with adaptive everyday functioning (Loe, Chatav, & Alduncin, 2015), but the clinical subscales may not clearly link to corresponding performance-based tools.

The primary aim of the current study was to investigate how well performance-based intelligence and EF measures predict EF everyday difficulties as reported by parents and teachers of four- to five-year-old preterm children. Due to a limited number of studies in this area, it is important to investigate the relationship between performance-based EF assessments and EF questionnaires of young preterm children in order to get a better understanding of which assessment tools best identify children at-risk of EF difficulties. Improved identification of EF difficulties in the preschool period can enable planning for supports and intervention as the children start schooling. Unlike some other studies of preterm children, which administered performance-based assessments and questionnaires concurrently, we wanted to understand how preschool assessment of EF predicted the EF after the children had entered kindergarten (i.e. the first year of formal schooling). Such research has a high value for practical clinical work where psychologists have to assess preterm children prior to start of the school to estimate their support needs. In Tasmania, Australia, where the study was conducted, children start kindergarten in the year following their fourth birthday (i.e., at four-to five years of age), which is younger than in many countries (Bertram & Pascal, 2002). This gave us an opportunity to investigate how well performance-based EF assessments prior to school predicted EF questionnaires when the preterm children enter school at a very young age.

We have previously published results confirming that our study group of four- to five-year-old preterm children displayed lower intelligence subtest scores and higher rates of impairment in performance-based EF when compared to a group of term-born peers (O'Meagher, Kemp, Norris, Anderson, & Skilbeck, 2017). The results of parent and teacher questionnaires were mixed. Parents in the preterm group and parents in the term group rated their children's EFs to be similar. The teachers of the preterm children reported the children as having more difficulties with inhibition, working memory, planning/organizational skills and self-control than did the teachers of the term group. Based on research to date (Garon, Piccinin, & Smith, 2016; Loe, Chatav, & Alduncin, 2015), we predicted that the level of agreement between intelligence test scores and reported EFs would only be low to moderate.

Methods

The preterm group participants were recruited from the routine follow-up of preterm infants offered by the Royal Hobart Hospital (RHH) Neonatal and Paediatric Intensive Care Unit (NPICU) Follow-up Clinic in Tasmania, Australia. The participants in this four-year-old assessment were 141 children (70 males, 71 females) (*M*=48.79 months, range 48-55 months). The mean gestational age at birth of the participants was 29.69 weeks' gestation (range 23.6-32.5 weeks). No children were excluded from the study, but a small number of participants could not be administered specific subtests due to significant global delay or sensorimotor issues (n=5). In these instances the participants were given the minimum score. There were no children with diagnosed syndromes in the cohort. The questionnaires were completed after the performance-based assessments. 82 parent and 105 teacher questionnaires were returned for the preterm children, and 49 parent and 46 teacher questionnaires for the term children. For missing data within the returned questionnaires, we used the multiple imputation method. Children in the term control group (*n*=77) were recruited from the local schools, pre-Kindergarten groups and word of mouth at the RHH. Children born at or over 37 weeks' gestation and with no diagnosed disabilities were eligible to participate. See Table 1 for the preterm group and term group characteristics. The preterm and term children were matched for age, sex and social risk.

Performance-based assessment (four years of age)

At four years of age, the children participated in performance-based intellectual (IQ) and EF assessment. The selected subtests of *Wechsler Preschool and Primary Scale of Intelligence, 3rd edition* (WPPSI-III) (Wechsler, 2002), namely Block Design, Matrix Reasoning, Information and Coding, were used to estimate general intelligence.

To assess executive functioning, subtests from the *Developmental Neuropsychological Assessment battery* (NEPSY-II; Korkman, Kirk, & Kemp, 2007) were administered to both groups (narrative memory recall, sentence recall and word generation), along with the *Shape School Task* (Espy, 1997), and the *Day-Night Stroop* (Gerstadt, Hong, & Diamond, 1994). The *Shape School Task* is a measure of inhibition, switching set and combination of both skills. It is a storybook-like assessment tool for preschoolers with human-like coloured shape figures. In condition A (Naming), the child is told that the figure's name is the colour, and the child has to say the name (colour) as quickly as possible without making any errors. In Condition B (Inhibit), the figures have both happy and sad faces. The child is told to name only the shape of the figures that are happy and inhibit saying the names of the sad shapes. In condition C (Switch), some figures are wearing hats and the child has to say the colour of the figures with hats and the shape for figures without hats, measuring cognitive shifting. Both conditions B and C require keeping two rules in mind, placing demands on working memory. The *Day-Night task* can be used with young children to measure switching and inhibition abilities. In this test, the child is required to say “day” when presented a page showing a night-time sky and “night” when shown a picture of a sun (16 trials). The WPPSI-III and NEPSY-II provide standardized age norms. For the Day-Night and Shape School tasks we used raw scores, as there are no standardized scores, but we age-controlled scores when comparing the preterm and term groups.

Questionnaire assessment (four to five years of age)

The year following the performance-based IQ and EF assessment, when the preterm children were four to five years old and had started kindergarten, the parents and kindergarten teachers were sent the *Behavior Rating Inventory of Executive Function–Preschool Version* (BRIEF-P) (Gioia et al., 2003) questionnaires relating to the preterm children’s EFs. The BRIEF-P is a rating scale developed to measure everyday behaviors associated with specific areas of executive functioning in children aged two to five years. The scales form three summary indexes: The Inhibitory Self-Control Index, the Flexibility Index and the Emergent Metacognition Index. There is also a Global Executive Composite combining metacognitive and emotional control scales.

The child’s socioeconomic risk was determined by using a composite social risk index (Roberts et al., 2008), which consists of six factors: family structure, education of the primary caregivers, occupation of the primary income earner, employment status of the primary income earner, language spoken at home and maternal age at birth of the preterm child. Each domain was scored on a three-point scale, with 0 representing low risk and 2 representing high risk, giving a total score between 0 and 12.

The study was approved by the Tasmanian Human Research Ethics Committee (H0011567) and the Tasmanian Social Science Ethics Committee (H0014174). This work was supported by the Royal Hobart Hospital Research Foundation under Grants 11-387 and 14-024. This study was a part of larger project, the results of which have been reported elsewhere (O'Meagher, Kemp, Norris, Anderson, & Skilbeck, 2017).

Results

Table 1 shows the participant characteristics. The preterm and term group were similar for sex, age, and overall social risk. Between-group comparisons (preterm vs. term) on standardized measures of intelligence and EF were conducted on all measures using independent-groups *t*-tests, with Bonferroni correction for multiple comparisons. As age standardized scores are not available for the Day-Night and Shape School tasks, analyses of covariance were performed, controlling for age. The Table 2 shows preterm group performed significantly more poorly than the term control group on all intelligence and EF scales (effect size, Hedges g=0.49 to 1.5). On BRIEF-P parental report, there were no significant group differences (g=0.00 to 0.24). However, the teachers reported elevated difficulties for the preterm group on several subscales: Inhibition, Working Memory, Planning/organizational Skills, Self-control and Overall Emergent Metacognitive skills (g=0.42 to 0.64).

Two separate simultaneous multiple regression analyses were performed to investigate associations between the parent and teacher questionnaires and 1) the WPPSI-III subscales (Block Design, Matrix Reasoning, Information and Coding) and 2) performance-based EF assessment results. Tables 3 and 4 show the associations for each of the subtests. The multiple regression analyses were conducted separately for the preterm and term groups. In the preterm group, WPPSI-III information was associated with several teacher BRIEF-P results (standardized β=-0.26 to -0.40; see Table 5) and WPPSI-III coding was associated with parent reported working memory on BRIEF-P (standardized β=-0.36). The Shape

School Switch efficiency was associated with BRIEF-P planning and organizational skills as reported by the parents (standardized β=-0.49). Day-Night efficiency was associated with teacher reported BRIEF-P emotional control (standardized β=-0.41) and NEPSY-II narrative memory recognition with teacher BRIEF-P planning and organizational skills (standardized β=-0.36). None of the other performance-based assessments had statistically significant associations with the parent or teacher reported EFs.

In the term group, WPPSI-III performance-based assessment results were not associated with any parent- or teacher-reported BRIEF-P EFs. Performance-based EF assessment tools of NEPSY-II word generation (standardized β=0.65 to 0.69), Day-Night efficiency (standardized β=0.44), Shape School Naming efficiency (standardized β=-0.41 to 0.45), NEPSY-II narrative recall (standardized β=-0.47 to 2.67) and narrative recognition (standardized β=0.41) predicted parent BRIEF-P scores to a varying degree. The Shape School Naming efficiency (standardized β=0.57), Shape School Switch efficiency (standardized β=0.51) and NEPSY-II sentence recall (standardized β=-0.59) were associated with some teacher BRIEF-P results.

Discussion

The main aim of the current study was to investigate how well performance-based intelligence subtests and EF assessments at the preschool age predict EF difficulties at home and school when preterm and term children start kindergarten. As we have reported previously, the preterm children had lower intelligence subtest scores and more performance-based EF difficulties than the term children (O'Meagher, Kemp, Norris, Anderson, & Skilbeck, 2017). Similar results have been consistently found in preterm groups of all ages (Anderson & Doyle, 2004; Mulder et al., 2009). The performance-based EF assessment results were not consistently associated with the parental and teacher reports of EF in everyday life for either preterm or term groups, in keeping with many other studies of general populations and older preterm groups (Conklin et al., 2008; McAuley et al., 2010; Sølsnes et al., 2014; Toplak et al., 2013).

Neuropsychological assessments evaluate an individual's cognitive strengths and weaknesses. Ideally psychometric assessments are associated with everyday living skills and behaviors and they have at least short-term predictive power. Despite this, EF assessment tools for preschool children remain scarce. Our research revealed that few of the performance-based EF assessment tools were associated with the EFs as reported by the parents and the teachers of four- to five-year-old preterm and term children. The performance-based EF assessment tools predicted parent and teacher reported EF more reliably for the term children, but still not consistently. It is somewhat concerning that the performance-based EF tests were not reliable at predicting the BRIEF-P results for the preterm children, as these tests may be used in clinical assessments when planning supports and funding for these children as they enter the school system. Thus, some children with real-life EF difficulties may be missed, whilst some children who were identified having EF difficulties at the assessment may not necessary display significant EF deficits at school and at home.

The WPPSI-III information subtest was a good predictor for many of the teacher BRIEF-P scales for the preterm children. The results of the information subtest may reflect a child's verbal intelligence and language skills and associated ability to, for example, follow instructions and comply with behavioral expectations in the classroom. The information subtest did not, however, predict the parental EF reports of the preterm group. Further studies are required to clarify the reasons behind this discrepancy. It is possible that instructions and vocabulary used in the classroom may differ from language used in a home environment, or the parents may be able to compensate their child's language difficulties by, for example, using gesture or more familiar sayings. Thus, better verbal skills may not be as important for successful EF at home when compared to school. Performance on the coding subtest was associated with parental reports of working memory, which is not surprising given coding is not a pure measure of motor speed but also taps into working memory. These patterns of information and coding predicting EF were nevertheless not seen for the term group. The reasons are not clear from the current literature and further investigation is needed relating to underlying processes that may connect different areas of intellectual functioning to reported EF in preterm vs. term groups.

Overall, the results indicate that performance-based tests that should predict EF in everyday life do not do so, or at least not very well. For example, it could be expected that subtests such as sentence recall are associated with reported working memory, as sentence recall has been created as a measure of working memory for the NEPSY-II assessment battery (Korkman, Kirk, & Kemp, 2007) and used in working memory studies (for example, Archibald & Joanisse, 2009). We did not find such associations in the current study. On the other hand, it is not clear if the BRIEF-P questionnaire's subscales reflect specific EF components, such as working memory, or whether BRIEF-P is just a measure of overall EF capacity (Fuhs & Day, 2011). It is also possible that parents and teachers completing the questionnaires may not identify subtle EF difficulties that are seen at performance-based EF assessments. Nevertheless, it is important to acknowledge limitations of EF assessment tools when making clinical judgement of children's EF, as single tests or subscales may not necessarily directly measure functions they try to tap into. This is not to say that the current EF assessment tools are not valuable, but clinicians have to be mindful that they are imperfect measures of EF. This suggests that further investigation of sensitivity and specificity of individual EF tests in relation to identification of EF difficulties in young children is required. More ecologically valid assessment tools may need to be developed to enable more reliable assessments at the preschool age.

A possible explanation for the observed discrepancies between EF testing in clinic settings and real-life measures has been offered by Barkley (2013), who has stated that performance-based EF assessments measure mostly the basic, momentarily, and instrumental EFs, but EF questionnaires are better at capturing the more complex adaptive, and strategic EFs, which enable us to navigate life in the long term. Thus, the BRIEF-P and other EF questionnaires tap into the ‘hot’ aspects of EFs, such as inhibition, while the performance-based EF tests may assess more ‘cool’ EFs, such as working memory (Anderson et al., 2010; Barkley, 2013; Payne, Hyman, Shores, & North, 2011).

There are many challenges in relation to the ecological validity of children’s cognitive assessments. These include methodological difficulties translating test performance into real-world ability, methodology of measuring real-world functioning, developmental factors, and intervening variables present in the everyday environment, such as, quality and expertise of the teacher (Johnson, Gilmore, Gallimore, Jaekel, & Wolke, 2015), supports the child may have in a classroom or stressors at home (Silver, 2000). Toplak et al. (2013) have noted that both performance-based and rating scale measures of EF are useful, albeit not interchangeable. Performance-based measures tap into the processing efficiency of cognitive abilities in structured settings and rating scales give information about individual goal pursuit in unstructured conditions and involve executive control. Thus, while there is a need to develop more ecologically valid EF assessment tools, the gap between performance-based assessments and questionnaire results may always persist. Greater caution in interpreting EF assessment results may be required to make valid assumptions of children’s EF solely based on a few assessment tool results.

Our study included, by design, a delay between performance-based assessment and the questionnaire of up to 18 months. It is possible that there may have been changes in the children’s EFs within this time, which could explain the reported differences between the performance-based assessment and questionnaire results. The fact that the term group had stronger associations between the performance-based assessment than the preterm group may also be explained by the fact that the term group's assessments and questionnaires were completed within a shorter time lag than those of the preterm group, although the relation between EF and chronological age may not be linear. A study design evaluating the associations between the performance-based assessments and questionnaires completed at the time of the assessment could provide further information about how closely the two methods are related. Nevertheless, ideally, clinicians would be able to use preschool EF assessment to identify those preterm children most at risk of having EF difficulties when they enter school. This early identification would help to enable intervention and support prior to the beginning of schooling. From that perspective, our study design may better reflect what is commonly happening in the real-world practice, that is, clinicians relying on the performance-based EF assessment tools to prepare their recommendations sometimes months before children start school. Adding EF questionnaires to the assessment battery at the preschool age could provide further information regarding the extent and the nature of preterm children's EF difficulties. Considering the time burdens of extending assessments, it should be verified what combination of performance-based and questionnaire tools adds most value to the assessment process. Further studies investigating how well EF questionnaires at preschool predict children's functioning as they start school are therefore required. It may be more cost effective to just utilise EF questionnaires, such as BRIEF-P, than assess children with time-consuming performance-based assessment tools. The present results are consistent with previous findings that the questionnaire results provided by different informants do not always agree (e.g., De Los Reyes & Kazdin, 2005). This leads to the recommendation that when assessing preschoolers, questionnaires designed for both parents and child-carers/pre-kindergarten teachers should be used. Further, we would recommend that that psychologists be trained to carefully analyse and interpret results from several sources to increase the validity of assessments of preterm children as they enter school. These assessments would ideally involve combination of questionnaires, observation and interviews (Anderson, 2002; Rabin et al., 2007), as well as performance-based assessments. This may mean more training resources and financial investment to make sure that psychologists have the necessary skills and knowledge, and also the time, to combine all the information when making conclusions about young preterm children's EF difficulties and their support needs. Correctly targeted early intervention, remediation and support prior to school entry could potentially improve educational and academic attainment of preterm children, who currently still lag behind their term peers in these areas.

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Disclosure statement

The authors report no conflict of interest.

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Tables

Table 1 *Preterm and Term Group Characteristics and Inter-Group Differences*

	Preterm		Term		*t*
Boys, *n* (%)	70 (49.6)		45 (58.4)		1.55
Girls, *n* (%)	71 (50.4)		32 (41.6)		1.55
Age (months)at EF assessment (mean, range) [a]	49.10 (48-58)		54.86 (48-67)		11.40***
Age (months) at parent questionnaires (mean, range)	58.33 (48-66)		57.35 (48-64)		-2.18
Age (months) at teacher questionnaires (mean, range)	58.40 (48-68)		58.41 (48-68)		-0.41
	M	*SD*	*M*	*SD*	*t*
Social risk index (the below risks combined)	2.98	2.61	3.26	2.62	0.74
Maternal age [b]	0.17	0.38	0.03	0.16	-3.94***
Family structure[b]	0.36	0.75	0.42	0.71	0.54
Main carer education level[b]	1.03	0.75	0.90	0.82	-1.20
Main income earner occupation[b]	1.01	0.91	1.03	0.85	0.14
Main income earner work status [b]	0.63	0.85	0.81	0.81	1.42
Language spoken at home [b]	0.07	0.12	0.07	0.34	1.57

[a] mainly scaled scores adjusted by age (utilised for comparison)
[b]social risk scores scaled 0(low)-2(high), e.g. maternal age <18 years=2, 18-21years=1, >21years=0

Table 2 *Intelligence and Executive Function Assessment Preterm and Term Groups*

	Preterm (*n*=141)		Term (*n*=77)					
	M	SD	M	SD	*t/F**	*p*	*df*	*g*
WPPSI-III								
Block Design[a]	8.11	3.17	10.55	3.10	4.82	<.001	184	0.77
Information[a]	9.05	4.10	11.05	2.64	3.96	<.001	152	0.54
Matrix Reasoning[a]	8.47	3.17	10.46	2.91	4.03	<.001	184	0.64
Coding[a]	7.05	3.55	10.95	2.37	8.72	<.001	149	1.20
NEPSY-II								
Narrative Recall[a]	5.23	2.33	8.77	3.25	8.17	<.001	170	1.35
Sentence Recall[a]	7.21	3.86	11.25	2.90	7.40	<.001	129	1.12
Word Generation[a]	8.04	3.38	12.07	2.34	9.08	<.001	140	1.29
Day-Night Efficiency[b]	0.18	0.12	0.37	0.15	8.60	<.001	136	1.50
(adjusted by age)	0.18	0.02	0.37	0.02	37.2	<.001	136	0.95
Shape A Naming Efficiency[b]	0.55	0.24	0.78	0.28	5.30	<.001	145	0.49
(adjusted by age)	0.58	0.03	0.73	0.04	6.15	.014	145	0.44
Shape B Inhibition Efficiency[b]	0.44	0.22	0.66	0.30	4.73	<.001	135	0.88
(adjusted by age)	0.46	0.03	0.63	0.04	8.67	.004	135	0.50
Shape C Switching Efficiency[b]	0.18	0.09	0.28	0.12	5.00	<.001	116	0.92
(adjusted by age)	0.19	0.01	0.28	0.02	12.2	.001	116	0.59

**F*-values provided for age-adjusted scores, analysed by ANCOVA

[a]scaled scores, [b]raw scores

Table 3 *Parent and Teacher Reporting of Executive Functioning (BRIEF-P, scaled scores) Preterm and Term Groups*

	Preterm M	SD	Term M	SD	*t*	*p*	*df*	*g*
Parents	(*n*=81)		(*n*=49)					
Inhibit	52.50	12.06	51.00	11.37	-0.71	.478	128	0.13
Shift	49.63	11.15	47.98	8.62	-0.89	.376	128	0.16
Emotional Control	50.90	12.69	50.65	11.69	-0.11	.912	128	0.02
Working Memory	55.48	15.61	52.04	11.68	-1.33	.155	122	0.24
Plan/Organize	51.62	13.97	51.61	13.12	-0.00	.998	128	0.00
Self-Control	51.88	13.18	50.04	11.07	-0.82	.416	128	0.15
Flexibility	49.99	12.62	49.06	10.69	-0.43	.669	128	0.08
Emergent Metacognitive	54.23	15.64	51.61	12.38	-1.06	.293	119	0.18
Teachers	(*n*=105)		(*n*=46)					
Inhibit	50.92	11.89	45.43	8.80	-2.81	.006	149	0.50
Shift	47.48	9.11	45.83	7.01	-1.09	.276	149	0.19
Emotional Control	46.15	7.31	45.46	7.39	-0.54	.592	149	0.09
Working Memory	54.09	12.34	47.00	7.57	-4.32	<.001	132	0.64
Plan/Organize	52.92	14.13	45.20	8.76	-4.09	<.001	132	0.60
Self-Control	48.86	10.14	44.65	9.49	-2.39	.018	149	0.42
Flexibility	46.92	8.57	44.83	7.35	-1.44	.151	149	0.25
Emergent Metacognitive	53.87	13.50	46.11	7.95	-4.40	<.001	136	0.64

Table 4 *IQ and EF Assessment Predictors for Reported EF for Preterm Children*

(*Regression model 1 IQ Assessment Results, Regression Model 2 EF Assessment Results)*

		b	Lower limit 95%CI	Upper limit	β	R^2
Parent questionnaires						
IQ assessment	*Associated EF questionnaire subscale*					
WPPSI-III-Coding	Inhibition	-0.79	-01.41	-0.16	-0.36*	0.21
EF assessment						
Shape School-C Switching	Planning/organizational	-16.12	-31.96	-0.29	-0.49*	0.20
Teacher questionnaires						
IQ assessment						
WPPSI-III-Information	Inhibition	-0.51	-1.0	-0.02	-0.26*	0.16
WPPSI-III-Information	Emotional Control	-0.25	-0.47	-0.03	-0.30*	0.11
WPPSI-III-Information	Working Memory	-0.80	-1.29	-0.29	-0.38**	0.25
WPPSI-III-Information	Planning/organizational	-0.53	-0.85	-0.21	-0.40***	0.27
EF assessment						
Day-Night Efficiency	Emotional Control	-6.37	-12.40	-0.35	-0.41*	0.22
NEPSY-II - Narrative Recognition	Planning/organizational	-0.47	-0.93	-0.02	-0.36*	0.17

*p<.05, **p<.01, ***p<.001

Table 5 *IQ and EF Assessment Predictors for Reported EF for Term Children*

(*Regression model 1 IQ Assessment Results, Regression Model 2 EF Assessment Results)*

		b	Lower limit 95%CI	Upper limit	β	R^2
Parent questionnaires						
EF assessment	*Associated EF questionnaire subscale*					
NEPSY-II-Word Generation	Inhibition	1.52	0.26	2.79	0.65*	0.32
NEPSY-II-Narrative Recall	Shifting	-0.49	-0.87	-0.11	-2.67*	0.20
NEPSY-II-Narrative Recall	Emotional Control	-0.54	-0.98	-0.11	-0.49*	0.49
NEPSY-II-Narrative Recognition	Emotional Control	0.76	0.14	1.38	0.47*	0.49
Shape School-A Naming	Emotional Control	-5.57	-10.63	-0.51	-0.45*	0.49
Day-Night Efficiency	Emotional Control	-10.71	-19.32	-2.10	-0.44*	0.49
NEPSY-II-Word Generation	Working Memory	1.52	0.49	2.55	0.69**	0.48
Day-Night Efficiency	Working Memory	-15.59	-28.40	-2.78	-0.44*	0.48
Shape School-A Naming	Working Memory	7.59	0.06	15.12	0.41*	0.48
NEPSY-II-Narrative Recognition	Working Memory	0.97	0.06	1.89	0.41*	0.48
Teacher questionnaires						
EF assessment						
Shape School-A Naming	Inhibition	13.72	2.57	24.88	0.57*	0.51
NEPSY-II -	Shifting	-0.64	-1.26	-0.02	-0.59*	0.42

Sentence Recall						
Shape School-C Switching	Emotional Control	18.24	0.88	35.60	0.51*	0.35

*p<.05, **p<.01, ***p<.001

Chapter 4. Congruency between parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten.

Congruency between parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten

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Congruency between parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten

Abstract: Parents of 82 and kindergarten teachers of 105 preterm (<33 weeks' gestation) and parents of 49 and kindergarten teachers of 46 term four- to five-year-old children completed executive function (EF) and behavior questionnaires. The preterm children were rated to have more EF difficulties than the term children by parents and teachers. On the behavior scales, the preterm children were reported as having more attention control difficulties than the term group, but no other behavioral problems. The parents reported higher levels of EF and behavioral difficulties than the teachers when both child groups were combined. The overall interrater reliability between parents and teachers in terms of children being in the clinical vs. non-clinical range for EF and behavioral problems was poor for the preterm and term groups. Conclusion: Based on this study, young preterm children need EF supports when commencing kindergarten. Noncongruent parent and teacher reporting nevertheless makes it challenging to identify the preterm children most at need of such supports. Further studies are needed to determine the factors impacting on reporting patterns, and also the best combination of EF and behavior assessment tools.

Keywords: executive function questionnaires; preterm; parent; teacher; behaviour; children

Introduction

Executive functions (EF) are adaptive, goal-directed behaviors that enable people to override more automatic thoughts and responses (Lezak, 1983). Executive functions include mental set shifting, working memory and inhibition. Shifting refers to the ability to change between mental sets and tasks, working memory consists of holding and manipulating multiple pieces of information in the mind, and inhibition enables overriding of more dominant responses (Miyake, Friedman, Emerson, Witzki, & Howerter, 2000). Executive functions undergo rapid changes in children from the age of three to five years, reflecting development of the attentional system and its connections to other brain areas (Garon, Bryson, & Smith, 2008), and continue to become more efficient as children mature (Carlson, 2005).

Children born preterm (<37 weeks' gestation) are at risk of having EF and behavioral problems, with the prevalence of overall neurobehavioral problems being up to 50% for very preterm children (e.g., Anderson & Doyle, 2004; Taylor, Klein, Drotar, Schluchter, & Hack, 2006). Behavioral problems have been associated with poorer academic outcomes in young children, but EF difficulties, such as poor attentional control, working memory, and planning, may have an even greater negative effect on school performance than behavioural problems (Brock, Rimm-Kaufman, Nathanson, & Grimm, 2009; Thorell, 2007; Willoughby, Kupersmidt, Voegler-Lee, & Bryant, 2011). With such high prevalence of EF and behavioral difficulties among preterm children, and the significant impact that these problems can have on the children's progress, it is important to assess both EFs and behavior of preterm children as they enter formal schooling. In this paper, we examine EF and behavioral difficulties using parent and teacher questionnaires in four- to five-year-old preterm children at the commencement of formal schooling.

In general, it has been recommended that preschool children, and especially those at risk of having developmental problems, be screened for any behavioral and cognitive difficulties (Burakevych, McKinlay, Alsweiler, Wouldes, & Harding, 2016; Doyle et al, 2014; EFCNI, 2011). However, there are no clear guidelines as to how often or at what age the children should be screened, or which tools to use, especially in relation to EFs (American Academy of Pediatrics, 2001; Poulou, 2013). There have been numerous studies of EFs and behavior in the older (over six years) school-age preterm population, but fewer focusing on executive functioning and behaviour of younger preschool preterm children. In the few studies that have examined preschool and early school-age preterm children, behavioral and EF difficulties have been detected (Aarnoudse-Moens, Smidts, Oosterlaan, Duivenvoorden, & Weisglas-Kuperus, 2009; Arpi & Ferrari, 2013; Loe, Chatav, & Alduncin, 2015). However, it should be noted that few studies have specifically investigated the four- to five-year-old age group, the age when children are entering school in some countries and states, including in Tasmania, Australia, where the current study was conducted.

Executive functions and behavior can be assessed by using direct observation, performance-based assessment tools or questionnaires completed by individuals who know the child; usually parents, caregivers and teachers. Questionnaires can provide valuable information about children’s behavior and EFs in everyday situations. Nevertheless, teacher and parent reports of children’s behavior have been reported to differ at both preschool and schoo- age, across different cultures, and when using different types of behavioral questionnaires (Achenbach, McConaughy, & Howell,1987; Berg-Nielsen, Solheim, Belsky, & Wichstrom, 2012; Korsch & Petermann, 2013; Markovic, Rescorla, Okanovic, Maraš, Bukurov, & Sekulic, 2016; Rescorla et al., 2014). In the studies that have been conducted with school-age children born preterm, the patterns of behavior reported by parents and teachers have been somewhat mixed. A meta-analysis of nine such studies suggested that the parent and teacher ratings of internalizing, externalizing behavioral and attention problems did not differ significantly (Aarnoudse-Moens, Weisglas-Kuperus, van Goudoever, &

Oosterlaan, 2009). However, not all studies fit this pattern. For example, other researchers have reported low levels ratings of agreement between parent and teacher ratings of attention deficit hyperactivity disorder ADHD (Leviton et al., 2017) and behavior (Scott et al., 2012) of extremely preterm children.

In a comprehensive analysis of the literature, De Los Reyes and Kazdin (2005) found that differences in reports of children's behavior in general population could not be explained by the child's gender, age, problem type, or social desirability. Also, parental or family characteristics, such social economic status or stress, could not directly explain the discrepancies. Instead, differences in informant reports can be explained in terms of trait effects, source effects, or a combination of both (Gomez, Burns, Walsh, & de Moura, 2003). Trait effects refer to the consistency of a child's behavioral presentation across informants within the same setting (Gomez et al., 2003). Poor trait effects occur if a child's behavior is perceived differently within the same environment by different people – for example, differences between mother and father reporting of a child's hyperactivity. Source effects are the biases that the informants may have. They could be true differences in the child's behavior in different situations (for example, a child is only oppositional at home), be informant-related (for example, a depressed parent may be overly negative about their child), or they could reflect both. Thus, when there are reporting differences, clinicians and researchers should consider both source and trait effects in explaining such discrepancies.

In the state of Tasmania (Australia), where our research was conducted, children can start kindergarten in the beginning of the calendar year following their fourth birthday. Although kindergarten is non-compulsory, about 98% of Tasmanian children attend kindergarten for the year prior to compulsory schooling (Australian Bureau of Statistics, 2016). At the international level, the age of kindergarten entry in Tasmania is relatively young, giving us a unique opportunity to investigate how the children's EFs and behavior are rated by both parents and school teachers at such a young age. Identifying preterm children at risk of having EF and behavioral difficulties at the beginning of schooling enables access to remediation and support for the children, families, and teachers. Due to often limited educational and therapeutic resources, it is essential to target those children who are most at risk of difficulties for intervention. Clinically, it is also important to evaluate different assessment tools, such EF and behavioral questionnaires, to find out how reliable they are in assisting this process.

The first aim of the current study was to compare the reported EF and behavioral difficulties between a four- to five-year-old preterm group and a term comparison group at kindergarten. Based on the previous studies of school-age children (e.g., Anderson & Doyle, 2004; Mulder, Pitchford, Hagger, & Marlow, 2009), we hypothesized that the parents and teachers would report greater EF and behavioral problems in the preterm group than the term group. We also examined the level of agreement between parent and teacher reports of preterm and term children's EFs and behavior at kindergarten. We expected parents to report higher levels of internalized behavioral problems, but not necessarily externalized and EF problems, than the teachers, for both groups. We based this expectation on the fact that internalized problems, such as anxiety, can be difficult to recognize in a busy classroom (Kolko & Kazdin, 1993). In contrast, we hypothesized that the teachers would report more EF problems than parents. EF difficulties may not be so evident in four- to five-year-olds at home, but teachers – especially those with extensive experience – could be more perceptive of mild EF difficulties in the classroom, where children may need to be more focussed and organized. Our second aim was to assess whether the parents' and teachers' opinions differ on which preterm children are identified as being at-risk for EF and behavioral difficulties. We predicted that the parents and teachers would classify preterm children differently when it came to clinical vs. nonclinical range in behavioral and EF problems, as has been the case for the classification of school-age preterm children (e.g., Leviton et al., 2017).

Methods

An invitation to attend a routine hospital-based assessment was issued to 184 children born preterm at less than 33 week's gestation, between 2007 and 2009. The Royal Hobart Hospital (RHH) Neonatal Intensive Care Unit Follow-up Clinic offered this routine preschool performance-based cognitive, as close as possible to the children's fourth birthday. Of the invited children, 141 (77%) participated in the study; 10 families were not contactable or had moved interstate or overseas, six declined to participate, and 27 did not attend after multiple follow-up invitations. The mean gestational age of the participating preterm children was 29.69 weeks (range 23.6-32.5 weeks), and none had congenital syndromes. Of the participating children, 38 were born <28 weeks' (27%), 79 between 28-31 weeks' (56%) and 24 between 32-33 weeks' gestation (17%).

The year following the initial performance-based cognitive assessment, when the preterm children were four to five years old and had started kindergarten (i.e., prior to compulsory formal schooling), their main carer (a parent) and kindergarten teacher were sent behavioral and EF questionnaires. Parents returned 82 and the teachers 105 questionnaires. The somewhat unusual pattern of teachers returning a higher rate of questionnaires than the parents may be explained by the fact that there was a longer time lag between the parent questionnaires being sent after consenting to the study than the teacher questionnaires.

The four- to five-year-old term comparison group participants (*N*=77) were recruited from local kindergartens, and by advertising at the RHH. Children born at or over 38 weeks' gestation and with no diagnosed disabilities were eligible to participate as controls. The children's main carer (parent) and teacher were provided the same behavioral and EF questionnaires to complete as the parents and teachers of the preterm children. For term children, overall 49 parent and 46 teacher questionnaires were completed. For missing data in the returned questionnaires, we used the multiple imputation method.

Overall social risk for the children was calculated combining six social risk factors identified by Roberts et al. (2008): family structure, education of primary caregiver, occupation, and employment status of primary income earner, language spoken at home and maternal age at the birth of the child. Each domain was scored on a three-point scale, with 0 representing low risk and 2 representing high risk, giving a total score between 0 and 12.

This study was approved by the Tasmanian Human Research Ethics Committee (H0011567) and the Tasmanian Social Science Ethics Committee (H0014174). It was supported by Starter and Clinical Research grants from the Royal Hobart Hospital Research Foundation research grant scheme. This study was a part of a larger project investigating preterm outcomes, the results of which have been reported elsewhere (O'Meagher, Kemp, Norris, Anderson, & Skilbeck, 2017; O'Meagher, Norris, Kemp, & Anderson, 2018).

Executive functioning was evaluated via the *Behavior Rating Inventory of Executive Function–Preschool Version* (BRIEF-P) (Gioia, Espy, & Isquith, 2003). BRIEF-P is a rating scale developed to measure everyday behaviors associated with specific areas of executive functioning in children aged two to five years. It consists of a rating form designed to be completed by parents, teachers, or other caregivers, with 63 items in five non-overlapping subscales: inhibition, shifting, emotional control, working memory and planning/organisation.

Behavior problems were evaluated via the two preschool parts of the *Achenbach System of Empirically Based Assessment (ASEBA),* (Achenbach, 1997): the Child Behavior Checklist for ages of one-and-a-half to five years (CBCL/1.5-5) was completed by parents, and the Caregiver-Teacher Report Form (C-TRF) was completed by kindergarten teachers. These are standardized behavior questionnaires for ages from 18 months to five years, and obtain parents' and teachers' ratings of 99 problem items. Seven syndrome scales (emotionally reactive, anxious/depressed, somatic complaints, withdrawn, sleep problems (CBCL only), attention problems and aggressive behavior) can be derived. The ASEBA DSM (Diagnostic and Statistical Manual of Mental Disorders)-oriented scales were not used, as the ASEBA DSM-oriented scales are not compliant with the current edition of this resource (DSM-5; American Psychiatric Association, 2013). Appendix 1 contains the BRIEF-P and ASEBA scales distribution and clinical cut-off scores.

Data Analysis

The statistical analyses were conducted with IBM SPSS Statistics versions 22.00 and 23.00 for Windows. We compared reported EF and behavior problems between different prematurity level groups (<28 weeks', 28-31 weeks' and 32-33 weeks' gestation), but the univariate analysis of variance showed no significant differences between groups in terms of these problems. Thus, all preterm children were treated as a single group for further analyses. In order to compare possible main effects and interactions of group (preterm vs. term) and informant (parent vs. teacher), a 2x2 analysis of variance was performed on scores for EF (BRIEF-P) and behavior (ASEBA). Bonferroni correction was made for multiple comparisons (adjusted alpha levels of .01 for BRIEF-P and .008 for ASEBA). We used T-scores in the comparisons. Higher scores on BRIEF-P and ASEBA indicate greater EF and behavior difficulties. An interrater reliability analysis using the kappa statistic was performed to determine level of agreement among the parent and teacher rating of EF (BRIEF-P) and behavior (ASEBA). A kappa of 1 indicates perfect agreement, while a kappa value of 0 indicates that interrater agreement is similar to chance. We reclassified child participants as being in the clinical range (T-score >65) vs. the nonclinical range (T-score <65) on their behavior and EF ratings for the kappa ratings. Effect sizes were measured as partial eta squares, which indicate the standardized difference between the means of the preterm and term groups and parent and teacher ratings.

Results

Table 1 shows the participant characteristics, the results of *t*-tests comparing the preterm and term groups, and their significance levels. As seen in the table, the preterm and term children were matched for age, sex, and overall social risk.

The BRIEF-P parent and teacher questionnaires consisted of five subscales (Cronbach's alpha α = .91 and α = .88 respectively). There were no significant informant by group interactions across any of the EF subscales. There was a significant main effect of group for all BRIEF-P subscales (inhibition $F(1,112)=5.81$, $p=.018$, $\eta_{pb}^2=.05$), shift ($F(1,112)= 4.10$, $p=.045$, $\eta_p^2=.03$), emotional control ($F(1,112)=9.47$, $p=.003$, $\eta_p^2=.08$) and working memory ($F(1,112)=6.62$, $p=.011$, $\eta_p^2=.06$), with the preterm group scoring higher across all domains (see Table 2 for the T-scores). There was a significant main effect of informant for inhibition ($F(1,112)=5.46$, $p=.021$, $\eta_p^2=.05$), working memory ($F(1,112)=7.63$, $p=.007$, $\eta_p^2=.06$) , and planning and organizational skills ($F(1,112)=4.70$, $p=.032$, $\eta_p^2=.04$) with parent ratings being higher than teacher ratings (see Table 2).

The ASEBA parent and teacher questionnaires consisted of six subscales (α= .86 and α= .72 respectively). For the behavioral questionnaires (ASEBA), no informant by group interactions were significant. There was a significant main effect of group for the attention problems scale ($F(1,108)=10.07$, $p<.001$, $\eta_p^2 = .08$), with the preterm children rating significantly higher than the term children (see Table 3 for the T-scores). There was a significant main effect of informant for emotional reactivity ($F(1,109)=16.88$, $p<.001$, $\eta_p^2=.13$), somatic complaints ($F(1,109)=33.38$, $p<.001$, $\eta_p^2=.23$), withdrawn/depressed ($F(1,109)=24.77$, $p<.001$, $\eta_p^2=.18$), and aggressive problems ($F(1,109)=4.93$, $p<.001$, $\eta_p^2=.04$) scales, with parental ratings being higher than teacher ratings (see Table 3)

The overall interrater reliability between the parents and teachers was found to be less than fair (κ < 0.21) for all EF questionnaire items. The interrater reliability was fair for attention problems (κ = 0.22) and withdrawal (κ = 0.32) in the behavioral ratings, with all the other behavioral rating agreements being less than fair (κ < 0.21). We also assessed interrater reliabilities separately for preterm and term children. Preterm children parents' and teachers' reporting (*n*=81) agreed at the fair level for attention (κ = 0.21) and withdrawal (κ =0.34), but less than fair (κ < 0.21) for the other ratings. For the term children (*n*=33), the level of agreement was less than fair (κ < 0.21) in all measures. The kappa results are detailed in Table 4. Since the mean scores of BRIEF-P and ASEBA for preterm and term children were not within the clinical range (T>65), the percentages of individual children falling within the clinical range are shown in Table 5.

Discussion

In the current study, parental and teacher reports of preterm and term kindergarten children's EFs and behavior were compared by using standardised rating tools. Overall, the preterm children were reported to exhibit more EF difficulties on the BRIEF-P than the term children when the parent and teacher ratings were combined. On the behavior scales (ASEBA), the preterm children were rated as having more problems than the term children in only one area; attention. The finding of preterm children having more EF and attention difficulties than the term group is consistent with the results of previous studies, especially for older school-age children (Anderson & Doyle, 2004; Mulder et al., 2009).

Contrary to our expectations, we found no differences between parental reporting on EF and behavior for preterm and term children. This result is in contrast to many other studies, which have found that the parents of preterm children tend to evaluate their children having more EF and behavioral issues than the parents of the term children (e.g., Arpi & Ferrari, 2013; Bhutta et al., 2002; Delobel-Ayoub et al., 2009; Johnson & Marlow, 2011;

Samara, Marlow, & Wolke, 2008). It has in fact been reported that the parents of preterm children may overestimate their children's difficulties (Estroff, Yando, Burke, & Snyder, 1994), and that the parents are biased in viewing their preterm children as vulnerable (Heinonen et al., 2013). However, it has also been suggested that the expectation that preterm children are relatively vulnerable may lower parental expectations, and thus lead to parents being less concerned about their children than would be expected (Schappin, Wijnroks, Uniken Venema, Jongmans, & Bruce, 2013). Additionally, parents may feel encouraged by the early developmental outcomes of their preterm children, who may have been given initially a cautious or negative prognosis.

One explanation for our preterm and term children's parents' ratings being similar may be that our groups were carefully matched for social risk, unlike in the majority of other preterm studies, which often have lower social risk or higher economic class families participating as comparison (Doyle et al., 2014). This may make our results more valid than those of some other studies. Also, all preterm children in our study could access regular state-provided medical/allied health surveillance and free preschool educational/allied health therapies. The impact of such follow-up on the outcomes of the preterm children has not been widely investigated. It is possible that the intervention our cohort of preterm children received improved their EF and behavioral outcomes as reported by their parents. It has been unclear how the EF and behaviour outcomes differ between children born more recently compared to the children born in the previous decades, as medical care of preterm children has fast improved in recent years. be Associations between medical care/early intervention and EF and behaviour outcomes for preterm children clearly warrant further investigation.

The parents in the current study rated higher levels of EF (inhibition, working memory, and planning and organizational) difficulties and behavioral (emotional reactivity, somatic complaints, withdrawn/depressed, and aggressive) problems than the teachers when preterm and term groups were combined. The overall level of agreement between the parent and teacher ratings of children being in the clinical vs. non-clinical range was not even moderately in agreement. Specifically, interrater reliability was only slight for EFs, and at best fair for behavioral difficulties, for both the preterm and the term groups. The overall interrater reliability between the parent and teacher ratings of children's EF being in the clinical vs. non-clinical range was also slight to fair for the behavioral difficulties for both the preterm and the term groups. Parental and teacher reporting of EF and behavior have been shown to differ in general populations, in many developmental and psychiatric conditions, and in school-age preterm children (e.g. Achenbach et al., 1987; Leviton el al., 2017; Shashi, Wray, Schoch, Curtiss, & Hooper, 2013). However, the evidence with the school-age preterm children has been mixed, with some reports indicating parents and teachers agreeing on behavioral difficulties (Aarnoudse-Moens, Weisglas-Kuperus et al., 2009). It is possible that the noncongruent reporting patterns between the parents and the teachers could be explained by reporter characteristics, differences in expectations of the children and the environments which the children are observed.

Clearly, parents and teachers do not consistently identify which children have significant EF or behavioral issues. These results raise an important question about the best clinical approach: how can clinicians determine which kindergarten children need more detailed assessment and support? Clinicians assessing young preterm children can find themselves in a position where they need to make conclusions and recommendations based on mixed reports from parents and teachers. One approach is to provide recommendations for support and treatment for the child in the specific environment where their EFs or behavior are seen to be problematic. This could be done by acknowledging reporting differences without giving the child a diagnosis or label, as suggested by De Los Reyes and Kazdin (2005). For a diagnosis of ADHD, for example, there needs to be an observable impairment in a child's functioning in at least two environments. An important aspect is to consider how much the reported EF and behavior issues impair the child's functioning (e.g., their ability to participate in everyday life activities, succeed in learning, and establish and maintain peer relations). It could be helpful to provide support strategies regardless of whether the reporting differences stem from the child's true internal factors, the environment, the reported characteristics of the children, or the combination of these.

More research is still needed to investigate what type of recommendations, if any, could support early school-age preterm children. Specific intervention programs have been trialed to improve young children's EF (Diamond, 2012; Diamond, Barnett, Thomas, & Munro, 2007) and behavioral control (Herd, Whittingham, Sanders, Colditz, & Boyd, 2014). While these behavioral programs have been reported to have positive impact in the short term, it has not yet been demonstrated that intervention programs can have a long-term impact on the cognitive outcomes of preterm children (Herd et al., 2014; Spittle, Orton, Anderson, Boyd, & Doyle, 2015). It has been suggested that intervention programs targeting EFs of preschool preterm children need to include both emotional and cognitive aspects, as emotional self-regulation difficulties may contribute to the lack of long-term success of such programs (Clancy, 2002). This may be due to emotional processes impacting highly on cortical activation of EFs, such as attention (Derryberry & Tucker, 1994).

While it has been shown that EF and externalizing (Schoemaker, Mulder, Deković, & Matthys, 2013) and internalizing behaviors are associated (Nelson et al., 2018), more studies are needed to investigate the associations between EF and behavior, especially relating to internalizing behavior and in children born preterm. Not much is known about the impact of moderating factors such as parental EFs and behavior on young preterm children's EFs and behavior. Nevertheless, higher social risk and especially lower parental education level have been shown to affect the development of preterm children's EFs (Aarnoudse-Moens, Weisglas-Kuperus et al., 2009; O'Meagher, Kemp, Norris, Anderson, & Skilbeck, 2017). Social risk negatively affects the behavioral outcomes of preterm children, and further, preterm children are at higher risk of developing behavioral problems than term children, even after controlling for social risk (Delobel-Ayoub et al., 2009; Spittle et al., 2009). Parenting style and affect has also been shown to have an impact on young preterm children's EFs. Specifically, intrusive, non-synchronized parenting style and negative parental affect have been associated with worse EF and behavioral outcomes (Clark & Woodward, 2014; Cueveas et al., 2014; Treyvaud et al., 2013). The impact of parental EF and behavior on young preterm children's executive functioning and behaviour needs to be further studied, to understand how much they affect preterm children's EF and behavior. It may be possible to target parenting styles or put other supports in place if there are clear associations between parental and child EF and behavior.

It will also be important to investigate the stability and predictive value of EF and behavioral problems of preterm children as they mature, as such studies do not yet exist (Arpi & Ferrari, 2013). While both young and older school-age preterm children have been shown to exhibit EF and behavior difficulties at the group level, it is less clear whether the same individuals with specific difficulties continue to experience these difficulties as they grow up. Some children may have EF or behavioral issues in primary school but their presentation may not differ from that of the general population as teenagers, while some other preterm children may develop EF and behavioral difficulties later in life. Also, the results from studies investigating how sex, medical risks and gestational age impact on EF and behavioral difficulties have been mixed (e.g., Arpi & Ferrari, 2013; Orchinik et al., 2011; Lundequist, Böhm, & Smedler, 2013; Pitchford, Johnson, Scerif, & Marlow, 2010; Potijk, Winter, Bos, Kerstjens, & Reijneveld, 2014), and more studies are needed to establish the associations, especially in young preterm children.

This study compared the perceived difficulties reported in the subjective opinions of parent and teachers. These opinions might be influenced by a range of informant characteristics, and future work could aim to determine whether and how informant characteristics such as sex, age, level of education and personality impact on how parents and teachers rate preterm children's behavior. Further research could also examine how other factors affecting the child participants, such as medical complications and the informants' knowledge of them, as well as the sex of the child, could affect how the informants viewed the child and interpreted their executive functioning and behavior.

Overall, this study further consolidates the findings that parents and teachers tend to evaluate children's EFs and behavior differently, not only in the general population, but also in children born preterm. The results point to the need for consensus on how different assessment tools, such as questionnaires, performance-based assessments and adaptive functioning scales, can be best utilised in planning supports and intervention for children born preterm. Furthermore, preterm children were reported by parents and teachers to have more EF – but not behavioural – difficulties than term children. Thus, EF supports for preterm children are needed when commencing kindergarten to reduce potential effects of EF difficulties on educational and academic attainment.

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Tables

Table 1 *Preterm and Term Group Characteristics and Inter-Group Differences*

	Preterm	Term	*t*	*g*
Boys, *n* (%)	70 (49.6)	45 (58.4)	1.55	0.16
Girls, *n* (%)	71 (50.4)	32 (41.6)	1.55	0.16
Age (months) at parent questionnaires (mean, range)	58.33 (48-66)	57.35 (48-64)	-2.18	0.22
Age (months) at teacher questionnaires (mean, range)	58.40 (48-68)	58.41 (48-68)	-0.41	0.23
	M (*SD*)	*M* (*SD*)	χ^2	*g*
Social risk index	2.98 (2.61)	3.26 (2.62)	0.29	0.11

t=*t*-test scores, **p*<.05, ***p*<.01, ****p*<.001

Table 2 *Main Effect of Group (Preterm and Term; 2x2 ANOVA)*

BRIEF-P T-scores	Preterm (*n*=81)		Term (*n*=33)	
	M	*SE*	*M*	*SE*
Inhibit	51.02	0.99	46.70	1.56
Shift	48.52	0.82	47.04	1.29
Emotional Control	48.61	0.85	47.62	1.34
Working Memory	54.38	1.21	48.17	1.90
Plan/Organize	51.91	1.20	47.06	1.88
ASEBA T-scores	**Preterm (*n*=78)**		**Term (*n*=33)**	
Emotional Reactivity	53.15	0.50	53.04	0.78
Anxious/Depressed	52.75	0.39	52.20	0.60
Somatic	52.88	0.47	52.32	0.72
Withdrawn	54.66	0.59	52.58	0.90
Attention Problems	55.66	0.69	51.63	1.06
Aggression	53.01	0.55	51.83	0.85

Table 3 *Main Effect of Informant (Parent and Teacher: 2x2 ANOVA)*

BRIEF-P T-scores	Parent (*n*=131)		Teacher(*n*=151)	
	M	*SE*	*M*	*SE*
Inhibit	50.33	1.20	47.38	1.01
Shift	48.92	1.05	46.64	0.84
Emotional Control	50.13	1.24	46.10	0.76
Working Memory	52.92	1.46	49.63	1.10
Plan/Organize	50.61	1.37	48.36	1.29
ASEBA T-scores	Parent (*n*=131)		Teacher (*n*=151)	
Emotional	54.62	0.76	51.58	0.36
Anxious/Depressed	53.01	0.54	51.94	0.37
Somatic	54.81	0.77	50.39	0.26
Withdrawn	55.39	0.81	51.84	0.42
Attention Problems	53.81	0.72	53.49	0.76
Aggression	53.33	0.83	51.52	0.39

Table 4 *Agreement Between Parent and Teacher Reports for Preterm and Term Groups*

Executive functioning (BRIEF-P)	κ	*SD*	*p*
Inhibit	0.16	0.07	.016*
Shift	0.13	0.08	.068
Emotional Control	0.06	0.08	.327
Working Memory	0.13	0.07	.037*
Plan/Organize	0.14	0.07	.048
Self-Control	0.12	0.07	.070
Flexibility	0.01	0.07	.845
Emergent Metacognitive	0.15	0.07	.021*
Behavior (ASEBA)	κ	*SD*	*p*
Emotional Reactivity	0.12	0.09	.009*
Anxious/Depressed	0.03	0.06	.648
Somatic Complaints	0.08	0.09	.110
Withdrawn	0.32	0.13	<.001***
Attention Problems	0.22	0.10	.006**
Aggression	0.14	0.11	.020*

*p<.05, **p<.01, ***p<.001

Table 5 *Percentages of Children Rated in Clinical Range (T>65) on ASEBA and BRIEF-P*

ASEBA	Preterm		Term		BRIEF P	Preterm		Term	
	Parent	Teacher	Parent	Teacher		Parent	Teacher	Parent	Teacher
Emotional Reactivity	6.1	1.9	12.2	0	Inhibit	13.6	11.4	6.1	6.5
Anxious/ Depressed	3.7	1.9	6.1	0	Shift	12.3	8.6	2.0	2.2
Somatic Complaints	8.5	1.9	8.2	0	Emotional Control	13.6	3.8	4.1	2.2
Withdrawn	11.0	6.7	8.2	0	Working Memory	22.2	21.0	12.2	6.5
Attention Problems	13.6	19.2	6.1	2.2	Plan/ Organize	17.3	18.1	12.2	6.5
Aggression	9.5	2.9	8.2	0					

Appendix 1 *Children's BRIEF-P and ASEBA Scales Distribution and Clinical Cut-off Scores (Rated by Parents and Teachers)*

Subscale	**Scales Distribution**	**Clinical Cut-off Score ($t \geq 65$)**
BRIEF-P boys parent inhibition	16-48	33
BRIEF-P boys parent shifting	10-30	21
BRIEF-P boys parent emotional control	10-30	20
BRIEF-P boys parent working memory	17-51	31
BRIEF-P boys parent planning/organisation	10-30	20
BRIEF-P girls parent inhibition	16-48	32
BRIEF-P girls parent shifting	10-30	20
BRIEF-P girls parent emotional control	10-30	22
BRIEF-P girls parent working memory	17-51	31
BRIEF-P girls parent planning/organisation	10-30	21
BRIEF-P boys teacher inhibition	16-48	35
BRIEF-P boys teacher shifting	10-30	20
BRIEF-P boys teacher emotional control	10-30	21
BRIEF-P boys teacher working memory	17-51	32
BRIEF-P boys teacher planning/organisation	10-30	19
BRIEF-P girls teacher inhibition	16-48	28
BRIEF-P girls teacher shifting	10-30	18
BRIEF-P girls teacher emotional control	10-30	19
BRIEF-P girls teacher working memory	17-51	30
BRIEF-P girls teacher planning/organisation	10-30	18
ASEBA parent (boys and girls) emotional reactivity	0-18	6
ASEBA parent (boys and girls) anxious/depressed	0-16	7
ASEBA parent (boys and girls) somatic	0-22	5
ASEBA parent (boys and girls) withdrawn	0-16	5
ASEBA parent (boys and girls) sleep problems	0-14	8
ASEBA parent (boys and girls) attention problems	0-10	6
ASEBA parent (boys and girls) aggression	0-38	21
ASEBA teacher boys emotional reactivity	0-14	5
ASEBA teacher boys anxious/depressed	0-16	6
ASEBA teacher boys somatic complaints	0-14	3
ASEBA teacher boys withdrawn	0-20	9
ASEBA teacher boys attention problems	0-18	11
ASEBA teacher boys aggression	0-50	22
ASEBA teacher girls emotional reactivity	0-14	5
ASEBA teacher girls anxious/depressed	0-16	7
ASEBA teacher girls somatic complaints	0-14	3
ASEBA teacher girls withdrawn	0-20	7
ASEBA teacher girls attention problems	0-18	9
ASEBA teacher girls aggression	0-50	19

Chapter 5. General Discussion

General Discussion

Summary of the Studies

Study Title	Aim	Methods	Results
Risk factors for executive function difficulties in preschool and early school-age preterm children	To establish if preterm children had more executive function difficulties than their term peers, and if so, to identify the social and perinatal risk factors associated with such discrepancy.	Measures: At four years: both groups assessed by using WPPSI-III (Block Design, Matrix Reasoning, Information and Coding), NEPSY-II (narrative memory recall, sentence recall and word generation), Shape School Task and the Day-Night Stroop At four to five years: Parents and teachers completed BRIEF-P questionnaires	-The preterm group performed more poorly on all measures of IQ and performance-based measures of executive function than the term group -The parental reports of executive function for preterm and term children did not significantly differ, but the teachers reported more executive function difficulties for the preterm group than the term group -Social risk, and especially the main carer education level, had strongest associations with outcomes
Examining the relationship between performance-based and questionnaire assessments of executive function in young preterm children: Implications for clinical practice	To determine whether specific performance-based executive function assessment tools were associated with executive functioning in everyday life as reported by parents and teachers of preterm and term children.	Measures: At four years: both groups assessed by using WPPSI-III (Block Design, Matrix Reasoning, Information and Coding), NEPSY-II (narrative memory recall, sentence recall and word generation), Shape School Task and the Day-Night Stroop At four to five years: Parents and teachers completed BRIEF-P questionnaires	-Intelligence and executive function assessment results did not have strong associations with the reported executive function difficulties

Congruency between parent and teacher reporting of executive function and behavioral difficulties in preterm and term children at kindergarten	To investigate the congruency of parent and teacher reporting of executive functions and behaviour of preterm and term children.	Measures: Parents and teachers completed BRIEF-P and ASEBA questionnaires when the children were four to five years old	-The parents reported higher levels of executive function and behavioural difficulties than the teachers for both preterm and term groups - The parent and teacher reports of behaviour and executive function were not in agreement relating to which children exhibited clinically significant executive function and behavioural problems

Overview of Thesis Aims and Outcomes

The current thesis explored executive function and behavioural outcomes of four- to five-year-old preterm children by comparing their performance-based assessment and questionnaire results to those of a group of children born at term. Additionally, this thesis examined executive function impairment related social and medical risk factors. Further, the thesis presents findings on congruency between performance-based assessments and reported executive function and parent and teacher reporting of children's executive functions and behaviour.

As expected, the preterm children demonstrated significantly lower performance in the IQ and executive function assessments than the term group. Similar results have been observed for school-age preterm children (e.g. Anderson & Doyle, 2004; Bhutta et al., 2002; Mulder et al., 2009) and for adults who were born preterm (Breeman, Jaekel, Baumann, Bartmann, & Wolke, 2015; Halmøy, Klungsøyr, Skjærven, & Haavik, 2012; Nosarti et al., 2007). In this way, findings of the current thesis reinforce the robust finding that the preterm population is vulnerable to executive function difficulties across different cohorts and age groups. There is a clearly a life-long risk of preterm individuals demonstrating executive dysfunction, which may hinder their academic, social and adaptive functioning. This is one of the few studies to use performance-based assessments with four- to five-year-old children on the cusp of entering formal schooling. Thus, the findings of this thesis address an important deficit in existing research literature regarding functioning in this age group and the impacts preterm birth have in this regard. There are not many studies like ours which have focussed on this critical age of four to five years when children's executive functions rapidly develop (Carlson & Moses, 2001; Diamond & Taylor, 1996; Lehto et al., 2003). Furthermore, the age of four to five years in Australia represents the age at which most children commence formal schooling, and thus executive functioning deficits may be more observable due to the demands of the school environment.

It is important to note that there are differences in the definition of "preschool" age across countries. Some previous "preschool" studies have been conducted with very young children (e.g., two-years-old or younger; e.g., Pozzetti et al., 2013), while others have included children up to the age of seven years (Aarnoudse-Moens et al., 2009; Baron et al., 2009). Some other studies of preschool preterm children, ranging from two to seven years of age, have reported poorer executive functioning for preterm than term children (e.g. Aarnoudse-Moens, Oosterlaan, Duivenvoorden, van Goudoever, &Weisglas-Kuperus, 2011; Baron et al., 2009; Clark & Woodward, 2010; Loe et al., 2015). Most of these studies have looked at only one or two executive functions, such as inhibition or working memory (e.g. Baron et al., 2009; Clark & Woodward, 2010), rather than use a whole battery of executive function assessment, as adopted within the current studies where a range of executive functions evident at preschool age (working memory, inhibition, shifting, emotional control, planning and organizational skills, mental flexibility and control, fluency) were assessed. By primarily using standardised and normed assessment tools, the results of the current series of studies are more generalisable than many of the pre-existing studies. Also, the results may be more familiar and useful to clinicians in guiding both assessment and intervention with preterm children than the newly developed laboratory-based assessments used in some studies (e.g., Baron et al., 2012). Furthermore, we assessed children born extremely preterm, but unlike like most studies, we also included very and moderately preterm children in our cohort, widening the base of our research and generalisability of findings. Our results that preterm children had more executive function difficulties than term children at preschool age further strengthens the premise that such difficulties can be detected already at such a young age. The findings also emphasise the need to develop assessment methods, supports and intervention to reduce the discrepancy in executive function outcomes between preterm and term children.

The role of social risk in preterm children's executive functioning development. We investigated social and medical risk factors for executive dysfunction to understand which young preterm children are most in need of developmental monitoring. To our knowledge, there have only been five articles published containing the keywords "risk factors", "executive functions" and "preterm", and these have all been published since 2013. This suggests there is a need to better understand which risk factors have significant impact on development of preterm children's executive function. Greater understanding of the role of risk factors in executive dysfunction can result in theory developments to guide interventions. Furthermore, we provided unique insights into the risk factors of executive function difficulties in young preschool children, as most of the studies relating to risk factors have included older individuals. Thus, our research into risk factors for executive functions of young preterm children is ground-breaking. We expected lower gestational age, higher social risk, male sex and medical complications to increase the odds of preterm children having executive function difficulties. The above risk factors have been shown to negatively impact on survival and cause major complications, such as cerebral palsy, for preterm children

(Beaino et al., 2010; Bhutta et al., 2002). Contrary to expectations, only social risk was strongly associated with performance across all executive function. Low educational level of the main caregiver (i.e., those without advanced tertiary, or post-high school education) was the strongest social predictor of poorer executive function in preterm children. We did not find strong associations between gestational age, hospital stay length (a proxy for medical complications) or sex of the preterm children with their executive function outcomes at age four to five years. One possible explanation of us not finding association between gestational age and EF outcomes may be that our preterm group consisted of not only children born extremely preterm but also very and moderately preterm. Thus, our preterm group may have been closer in their presentation to the term group and general population than in many other previous studies. Our finding regarding the importance of the social environment in predicting executive dysfunction is consistent with general population studies, which have shown that higher social risk backgrounds and lower parental education levels can negatively impact the development of children's executive functions (Hackman & Farah, 2009; Merz & McCall, 2011). Reasons behind this are still unclear but they could relate to either lower parental cognitive functioning or other reasons, such as stress, family dysfunction and lower financial resources, to support the development of a child. As such, there is a need to investigate the mechanisms underlying the observed poorer executive functions of preterm children living in higher social risk families. In addition to studying risk factors, it would also be important to study what are the possible resilience factors protecting at-risk preterm children from executive dysfunction. Furthermore, effective intervention programs are clearly needed to support young preterm children living in high risk environments.

Surveillance and assessment of preterm children's executive functions. For preterm children to access supports and intervention, the children needing such resources have to first be identified. How this identification process is best organized is unclear, as there are no generally accepted guidelines for clinicians working with young preterm children. There are position papers available for best assessment approaches in the adult population, especially related to dementia and traumatic brain injury (e.g. Diniz et al., 2008; Roebuck-Spencer et al., 2017). Advice and internationally approved position papers for the paediatric and especially for young preterm children are lacking, although an Australian working group (Doyle et al., 2014) has recommended that funding for neonatal intensive care should include a commitment to fund follow-up assessments for preterm children and other high-risk children at least until early school-age, a recommendation that results of the current studies would support, particularly in relation to executive functions. At the moment, high-risk preterm children may not get assessed until they start performing poorly academically or develop behavioural problems. Intervention at a later age may not be as effective as it could have been at earlier stages of development when the foundations of skills were laid (Doyle et al., 2014). The Doyle et al. working group provided general recommendations relating to following up preterm children and named several psychometric assessment tools that could be used to assess the children's cognition, behaviour and executive function, however most were appropriate only for school-age children aged six years and over (Doyle et al., 2014). Furthermore, there is a need to fine-tune the recommendations to investigate which of the assessment tools can form the most cost-effective, ecologically valid assessment battery to assess executive function difficulties of preterm children, especially before they enter schooling. Overall, such assessment tools are scarce for young children in general. We investigated if currently available performance-based intelligence and executive function assessment tool results at preschool age were predictive of preterm children's reported executive function difficulties when they entered kindergarten. We found that intelligence and executive function assessment results did not have strong associations with parent and teacher reported executive function difficulties for either preterm or term groups. One explanation is that the performance-based executive function and the questionnaire assessment evaluate different aspects of executive functions. Toplak and colleagues (2013) noted that both performance-based and rating scale measures of executive functioning are useful, albeit not interchangeable. The performance-based measures tap into the processing efficiency of cognitive abilities in a structured setting, whereas rating scales give information about individual goal pursuit in unstructured conditions and involve executive control. Also, performance-based executive function assessment tools are generally used in a structured and supported environment with few distractions. Thus, the performance-based assessment results may not detect the difficulties that may occur in real-life situations, which inherently have more distractions (McAuley, Chen, Goos, Schachar, & Crosbie, 2010). On the other hand, preterm children may be able to compensate for difficulties evident in performance-based assessments by using other skills and resources available in real-life settings (Heinonen et al., 2013). For example, preterm children may be assisted and reminded by parents and teachers in real life more than in the assessment situations, or the preterm children may be able to follow other children’s lead when given instructions, thus masking their executive function difficulties.

Another possible explanation for the observed discrepancies between performance-based executive function testing and real-life measures has been offered by Barkley (2013), who stated that performance-based executive function tests tend to assess rudimentary executive functions at a particular moment. In daily life, more adaptive, tactical and strategic executive functions are required to enable socially cooperative and reciprocal activities that can last for much longer, sometimes years.

The construct validity of performance-based executive function tasks and the questionnaires may explain why performance-based and questionnaire assessment results are not congruent. Performance-based executive function tasks have been shown to have fairly robust construct validity when detecting impairment vs. no impairment (Chan, Shum, Toulopoulou, & Chen, 2008), but different performance-based executive function tasks assess specific elements of executive function, and performance can vary according to modality, content and complexity of a task. This has clinical implications when selecting and interpreting executive function assessment tasks for preterm and other children, because children may do poorly in one test measuring executive function, such as working memory, but succeed in another. For example, the working memory task may be verbal or visual in modality, include content that may or not interest the child, or be more complex and include other executive function than working memory, such as switching, which may impact on the child's results. It is not unusual to see this variation and in order to confirm executive dysfunction in one area, such as working memory, deficiency should be found in more than one subtest of working memory (Lezak, 1995). Furthermore, some authors have suggested that in general the executive function rating scales are more ecologically valid than the performance-based assessment tools (Spooner & Pachana, 2006; Sølsnes, Skranes, Brubakk, & Løhaugen, 2014). On the other hand, some researchers have questioned the validity and utility of rating scales, such as the BRIEF-P, due to its factor structure. Duku and Vaillancourt (2014) reported that some of BRIEF-P subscales could be separated into further categories, such as the inhibition scale to "awareness" and "impulsivity" and the shifting subscale into "inflexibility," "adjusting," and "sensory", thus questioning the validity of the current scale structure. Spiegel, Lonigan, and Phillips (2016) confirmed that the factor analysis results of BRIEF-P were very mixed. Further investigation of sensitivity and specificity of individual executive function assessment tools in young children is required to address these identified equivocal results. More ecologically valid assessment tools may need to be developed to enable more reliable assessments of executive function at the preschool age. As indicated above, the current executive function assessment tools, performance-based and questionnaires, have limitations in general. There is a greater need to develop executive function measures for young (under six-year-old) children, as currently there very few assessment tools available. Test impurity is one of the challenges when creating assessments tools, as typically EF tests can measure multiple components of EF (Taylor & Clark, 2016), and it may also be difficult to exclude the impact latent factors may have on the assessment results (Stålnacke, Lundequist, Böhm, Forssberg, & Smedler, 2018). One alternative method to overcome these limitations may be to create experiments which manipulate test demands and have different levels of complexity (Taylor & Clark, 2016). Furthermore, it is important to create more ecologically valid assessment tools to increase the practical value of EF assessments (Alvarez, & Emory, 2006). Such assessments could include everyday tasks like planning a birthday party or a trip to an amusement park. Using virtual reality-based assessments offers one possible option to improve ecological validity of EF assessments (Jansari, Devlin, Agnew, Akesson, Murphy, & Leadbetter, 2014), although such tools need to still be further evaluated. Potentially young children may be more motivated in assessment situations where electronic devices, such as computers and tablets, are used, and the assessment results may not truly reflect real life situations where such devices are not present.

Our study looked at the predictive value of executive function assessments in relation to questionnaires, but there is also a need to evaluate the associations between the performance-based assessments and questionnaires completed at the time of the assessment. Nevertheless, before recommending adding executive function questionnaires to a test battery, it is important to know how well earlier administered executive function questionnaires at preschool predict children's functioning as they start school, as the psychology resources are limited in most countries and the existing staff resources are under pressure to spend their time efficiently (Goodheart, 2010; Institute of Medicine, 2015). Additionally, it is important to also investigate stability and predictive value of executive functions problems of preterm children as they mature. We know now that at the group level both preschool and school-age preterm children are at risk of having executive function difficulties. However, it is not known if some preterm children who have been identified as having executive function difficulties at preschool age will have executive function difficulties as they mature, or whether it is possible to overcome such problems. Also, it is possible that some school-age preterm children who have executive function difficulties did not have such issues when at preschool and instead developed them later. This is a plausible possibility, as executive functions undergo such a fundamental change prior to age of six to seven (Carlson, 2005; Garon et al., 2008). Thus, it would be important to study trends by following up the same group of children, as many longitudinal studies of preterm children's EF have used different age groups (different cohorts) and assessed if all the groups exhibit the same executive function problem, such as working memory deficit. This does not give us information if a group of children who have executive function difficulties at an earlier age continue having them as they mature: it is possible they outgrow their difficulties but another group develop them later. Also, different assessment tools have been used across different ages in many studies, so it possible they actually tap into different latent factors, such language skills and processing speed (Huizinga, Dolan, & van der Molen, 2006). Furthermore, most longitudinal studies have included only children over the age of five (Huizinga, Dolan, & van der Molen, 2006; Stålnacke, Lundequist, Böhm, Forssberg, & Smedler, 2018), overlooking an important age-group of four-year-olds at the time EF functions undergo rapid changes. These issues warrant further research and theoretical framing in order to develop guidelines to determine the age or ages at which preterm children's executive functions should be assessed.

Parent and teacher reports of children's behaviour and executive functions. The current thesis also explored the executive function and behaviour reporting patterns of parents and teachers. Against expectations, parental reports relating to executive function did not significantly differ between those with preterm versus term children. In contrast, teachers reported more executive function difficulties for the preterm children than the term children, specifically in the areas of inhibition, working memory, planning/organizational skills and self-control. Mixed results regarding parent and teacher reporting of executive functions have been reported in the general population (McCann, Rider, Weiss, Litman, & Baron, 2013; Pedersen, 2005; Rahbari & Vaillancourt, 2015) and in the preterm groups (Bhutta et al., 2002; Heinonen et al., 2013). Previous research, mainly in school-age and older preterm populations, has indicated that parents of preterm individuals tend to report more difficulties than parents of term individuals (Arpi & Ferrari, 2013; Bhutta et al., 2002; Delobel-Ayoub et al., 2009; Johnson & Marlow, 2011; Samara et al., 2008), although opposite results have also been found (Schappin et al., 2013). Our results indicate that the parents of preterm children do not necessarily report more executive function difficulties than the parents of term children. This result may have been due to the fact that executive function difficulties are not as evident in the home environment as at school. To our knowledge, ours is the first study investigating parent and teacher reporting of preterm children's executive functions at the age of four to five years. More studies are needed to establish how and why parent and teacher reporting of young preterm children's executive functions differ.

In our third study, we found that the parents of both the preterm and the term groups reported higher levels of executive function and behavioural difficulties than the teachers. The preterm children were reported to exhibit more executive function difficulties than the term children when the parent and teacher ratings were combined. On the behaviour scales, the preterm children were rated by the parents and the teachers as having more attention problems than the term children but no other behavioural issues. We expected parents of preterm children to report more behavioural issues than the parents of the term group based on preterm school-age child studies. Most previous research looking at executive function and behavioural outcomes has found that the parents tend to evaluate their preterm children having more difficulties than the parents of the term children (Arpi & Ferrari, 2013; Bhutta et al., 2002; Delobel-Ayoub et al., 2009; Johnson & Marlow, 2011; Samara et al., 2008). One possible explanation is that the parents of preterm children may have an expectation that preterm children are vulnerable, which may also lower parental expectations relating to the outcomes, and thus lead to parents being less concerned about their children than would be expected (Schappin et al., 2013). Additionally, parents may feel encouraged by the early developmental outcomes, for example, walking and talking, of their preterm children, who may have been given initially a cautious or negative prognosis. One possible explanation for the lack of difference in parental reporting between the preterm and term groups is that we matched the two groups for social risk, and thus our results contrasted with some other studies (Arpi & Ferrari, 2013; Bhutta et al., 2002; Delobel-Ayoub et al., 2009; Johnson & Marlow, 2011; Samara et al., 2008). Lower social risk families tend to sign up for research more readily than higher social risk families (Doyle et al., 2014), and children coming from lower social risk families have more proficient executive functions (Msall et al., 2007; Palfrey, 2006), which may explain why some other studies have found the differences in parent reporting of executive functions but we did not. However, it is possible that there was a selection bias for the term and/or the preterm group in our study. It would have been based on self-selection, as the parents of both preterm and term voluntarily chose to participate in this study.

In our study, the parent and teacher reports of executive function and behaviour of both preterm and term children differed significantly: parents and teachers were not in agreement about which children exhibited clinically significant executive function and behavioural problems. Parental and teacher reporting of executive function and behaviour have been shown to differ in general populations, in many developmental and psychiatric conditions, and in school-age preterm children (e.g. Achenbach et al., 1987; Leviton el al., 2017; Shashi, Wray, Schoch, Curtiss, & Hooper, 2013). However, the evidence from school-age preterm children has been mixed, with some reports indicating parents and teachers agreeing on behavioral difficulties (Aarnoudse-Moens, Weisglas-Kuperus, van Goudoever, & Oosterlaan, 2009), and others finding that the parental and teacher reports differed (Leviton el al., 2017; McCann et al., 2013). Our study indicates that there is more evidence for noncongruent than congruent reporting patterns, which may be due to reporter characteristics, such as education level, sex and age, differences in expectations of the children, or the environments which the children are observed in. These factors warrant further research. Also, the possible impact of informants' knowledge and perception of social risk factors, sex of the child, medical complications and gestational age on their reporting of executive functions and behaviour needs to be investigated, to evaluate the reliability of informant reports in the case the informant knows these factors. For example, teachers may overestimate difficulties if they know the child was born extremely preterm or they may have a biased view of how boys versus girls should behave. As questionnaire results provided by different informants can differ (De Los Reyes & Kazdin, 2005), this leads to the recommendation that when assessing preterm children, questionnaires designed for both parents and child carers/teachers should be used.

As indicated previously, there are no internationally recognized guidelines for surveillance of preterm children's executive function and behaviour outcomes. We do not know how and which assessment tools, such as questionnaires, performance-based assessments and adaptive functioning scales, can be best utilised in planning supports and intervention for children born preterm. It is important to make sure that clinicians assessing preterm children are provided with sufficient training in interpreting and combining assessment results, so they can make relevant recommendations to support preterm children's executive function development. These assessments would ideally involve combination of questionnaires, observation and interviews (Anderson, 2002; Rabin, Burton, & Barr, 2007), as well as performance-based assessments. While informant-rated questionnaires and performance-based measures of executive function are not strongly associated, both questionnaires and performance-based assessment tools may provide important information about executive functioning (Loe et al., 2015; Ritter et al., 2014). Getting information from multiple assessment sources may require more training resources and financial investment to make sure that psychologists have the necessary skills and knowledge, and also the time, to combine all the information when making conclusions about young preterm children's executive function difficulties and their support needs. Correctly targeted early intervention, remediation and support prior to school-entry could potentially improve educational and academic attainment of preterm children, who currently still lag behind their term peers in these areas.

Limitations

It is acknowledged that this series of studies had some limitations. We did not assess the intellectual and executive functioning of the parents of the children in the current study. Such assessment could clarify how much impact the genetically inherited intellectual capacity and the parents' executive function skills may have on the development of preterm children's executive functions compared to other social, medical and educational factors. We acknowledge that the reported results are based on subjective opinions of parents and teachers. We did not evaluate informant characteristics, but in future it would be useful to determine whether and how informant characteristics such as sex, age, level of education and personality impact on how parents and teachers rate preterm children's executive functions and behaviour. This may give clinicians further understanding of how such factors may affect reporting patterns of executive function and behaviour and encourage caution when interpreting assessment results. Such research could also prompt clinicians to seek further information if there is a reason why particular reports of executive function and behaviour may not be as reliable as others. Further research could also examine how other factors affecting the child participants, such as medical complications and the informants' knowledge of them, as well as the sex of the child, could affect how the informants viewed the child and interpreted their executive functioning and behaviour. Furthermore, in our studies we did not separately control the preterm children's IQ, like is some other studies. However, IQ overlaps with executive functioning, and thus it is not recommended that IQ be used as a co-variate, as it can produce overcorrected findings about neurocognitive function (Dennis, Franci, Cirino, Schachar, Barnes, & Fletcher, 2009). Another reason that we did not use IQ as a covariate is because there are good arguments that IQ is a sufficiently variable factor to make it an inappropriate covariate when considering specific paediatric risk groups, such as preterm children (Dennis et al., 2009).

The preterm children who participated in the study differed from the non-attendees, which needs be considered when evaluating the results. The preterm attendees had a lower gestational age and birthweight and had more medical complications than the non-attendees as shown in Table 2 on page 20. Thus, our study group may have been, on average, more disadvantaged than when considering the average profile of the whole cohort of preterm children born in Tasmania during the study period. While we acknowledge the challenges of matching the study groups by all variables with the whole cohort and the comparison group, it would be recommendable to attempt to do so.

A further limitation for our study was the significant difference in age between the preterm and term group at the performance-based assessment, with term children on an average being older than preterm group. Although we were not able to match the children's age at the time of the performance-based assessment (because we had to assess children according to the availability of the children and their families), we did match for age at the time of the questionnaire completion, as planned. Our aim was to assess all children on the parent and teacher questionnaires as the children entered kindergarten at about 58 months of age, which we achieved. Age-matching was also complicated by our important aim of matching children on social risk, as we wished to avoid the tendency of previous studies to have comparison children with lower social risk than preterm children. There are challenges in engaging high social risk families in research projects like ours (Doyle et al., 2014), so we valued achieving the goal of matching the preterm and term groups for social risk. Regardless, the problems of age-matching were mitigated by our use of standardised scores that control for age (WPPSI-III and NEPSY-II). While there are not yet standardised scores available for the Shape School and Day-Night tasks, they do offer age-expected scores for different age ranges. We conducted an additional statistical comparison utilising these scores and found that the preterm children's performance was poorer than the term children's, like our initial results. Thus, we concluded the results cannot be explained by the age difference alone.

Another issue is potential age-correction for prematurity, which has been applied in some preterm studies. We did not adjust the age of the preterm children for prematurity because our focus was on the more practical question of how the preterm children fare at school entry compared to their same-age peers. The school entry point is not usually lowered based on gestational age, and the preterm children are expected to cope similarly in the school environment compared to their peers. While it is known that the effects of prematurity on developmental and cognitive outcomes persist during school-age, our study had a strongly clinical focus. In clinical settings, the examiners who use assessment tools such as WPPSI-III and NEPSY-II do not routinely correct the child's age for prematurity beyond the first couple of years. Doyle and Anderson (2016) have recently called for an international policy relating to correcting for prematurity, but also for flexibility to deal with the different objectives of the assessments. In longitudinal studies it may be advisable to correct the age for prematurity also after three years of age, as otherwise it may be difficult to interpret any change in preterm children's functioning if the age was corrected at an earlier but not later assessment occasion (Wilson-Ching, Pascoe, Doyle, & Anderson, 2014). Furthermore, it may be more justifiable to perform age-corrections for children born extremely preterm rather than for those born later in gestation, as the difference an age-correction makes for the latter group is minimal (Wilson-Ching, Pascoe, Doyle, & Anderson, 2014). As our preterm group also included very and moderately preterm children, age-correction was not crucial. At the commencement of our study, the recommendations we found did not endorse using corrected scores for prematurity past three years of age (Engle & American Academy of Pediatrics Committee on Fetus and Newborn, 2004). In our opinion, we have compensated possible issues with the age-matching and age-correction satisfactorily.

Summary of Implications and Recommendations

Implications of this thesis
• There is a need to narrow the gap in intelligence and executive function outcomes between preterm and term children • More effective identification of executive function difficulties in preterm children prior to entering school is required to enable early supports • Social risk is malleable, unlike risk factors such as gestational age and sex, and it is possible to target social risk via intervention • There is more need for executive function than behavioural supports for preterm children when they enter schooling • There is discrepancy on how parents and teachers report executive functions of young preterm children, which complicates clinical decision making • Clinicians using executive function assessment tools need to apply caution when interpreting the results of the assessment of preterm children due discrepancies in performance-based and reported executive function results • It may be difficult for clinicians determine which kindergarten children need more detailed assessment and support due non-congruent assessment results and informant reports
Recommendations and further research
• Further research is needed about how sex, medical risks and gestational age impact on executive functions and behavioural difficulties of preterm children and how these may interact with the social risks identified within the current thesis • There is also a need to investigate why the children from families at greater social risk have poorer outcomes as this provides a target for intervention • Research focus could also be shifted to resilience factors, that is, what are the factors that protect at-risk children being spared from executive function difficulties • Need to study how parental executive functions and behaviour impact on young preterm children's executive functions and behaviour • May need to target parenting styles or put other supports in place if there are associations between parental and child executive functioning and behaviour • Intervention programs focused on reducing the impact of social risks to improve preterm children's executive function outcomes at early school-age are needed • This thesis has identified that it is preferable to use multiple informants when evaluating executive functions and behaviour. However, further studies relating to parent and teacher reporting of executive functions are required, for example, to evaluate informant and child characteristics which may influence questionnaire results • Further investigation is also required to determine how informants' knowledge and perception of social risk factors, sex of the child, medical risks and gestational age impact on their reporting of executive functions and behaviour • It is also important to look at the stability and predictive value of executive dysfunction and behavioural problems of preterm children as they mature, as it is possible that some children "outgrow' their early childhood difficulties while others may develop them later • There is a need for consensus on how different assessment tools, such as

questionnaires, performance-based assessments and adaptive functioning scales, can be best utilised in planning supports and intervention for children born preterm

- Executive function assessments would ideally involve a combination of questionnaires, observation and interviews. Psychologists need to be trained to carefully analyse and interpret results from several sources to increase the validity of assessments of preterm children
- More ecologically valid performance-based tests and/or executive function questionnaires may need to be developed for young children
- As our study looked at the predictive value of executive function assessments in relation to questionnaires, there is a need to evaluate the associations between the performance-based assessments and questionnaires completed at the time of the assessment. Before recommending adding executive function questionnaires to a test battery, we need to know how well earlier administered executive function questionnaires at preschool predict children's functioning as they start school

Conclusions

In this thesis we have shown preterm children are at risk of demonstrating executive function difficulties as young as four to five years. With limited resources in health and education, it is important to try to identify the high-risk preterm children for longer term follow-up. This task can be difficult, as development and outcome of executive functions can be influenced by several factors, such as gestational age, medical complications, socio-economic factors and sex of the child (Beaino et al., 2010; Bhutta et al., 2002; Hintz et al., 2006; Taylor et al., 2006). While decreasing gestational age is related to poorer overall outcome and increased number of impairments (Hirschberger et al., 2018), executive dysfunction may not be so linearly associated with gestation age, as shown in our study. Thus, the solution may not be to just set an arbitrary gestational age range for the preterm children whose development should be monitored. As we reported, social risk factors were the strongest predictors for executive function difficulties. Supports and programs to reduce social disadvantage of preterm children are needed to improve cognitive and academic outcomes (Spittle, Treyvaud, Lee, Anderson, & Doyle, 2018). Given the importance of the years of four to five for executive function development (Carlson, 2005; Garon et al., 2008), it is crucial that preterm children are identified early and prior to formal school. There is a further need for development of executive function assessment tools for young (under six-year-old) children, as the selection of available tools is limited. Also, as shown by our research, the current performance-based assessment tools and questionnaire do not clearly identify the preterm children at risk of executive function difficulties, who would need intervention and support to give them the best chance of successful life.

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Appendices

Appendix A. Abstract of: O'Meagher, S., Kemp, N., Norris, K., & Anderson, P. (2017). Sex differences in executive functions of preterm and term preschool children. Abstracts presented at the International Neuropsychological Society Mid-Year Congress 2017. *Journal of the International Neuropsychological Society, 23*(S2), I-59.

Sex differences in executive functions of preterm and term preschool children

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Objective: It is known that school-age children born preterm are at risk of having executive function difficulties. However, little is known about sex differences in executive functions at preschool age. The aim of this study was to investigate sex differences in the executive functioning of preterm and term preschoolers using performance-based executive function assessment tools.
Participants and Methods: 141 children born very preterm (< 33 weeks of gestation) and 77 term controls were assessed at the age of 4 years, prior to starting kindergarten, on standardized performance-based executive function tests (parts of the Developmental Neuropsychological Assessment-II (NEPSY-II), Day-Night Stroop and Shape School).
Results: The preterm children performed significantly more poorly on all executive function tests than did the term children. Overall, girls performed significantly better than boys on three of the seven tasks. There was no significant interaction between group and sex for any of the tasks. However, the pattern of differences between boys and girls was quite dissimilar for the preterm and term groups.
Conclusions: Preterm preschool children performed more poorly than term children on executive function assessments. Further, preterm females outperformed preterm males on some of these tasks, but these sex differences were no greater than those seen in the general population. Thus, the potential disadvantages of prematurity observed for preterm boys than for preterm girls were no greater than for children born at term. However, the pattern of ability for boys versus girls might differ from that in the general population.

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Appendix B. O'Meagher, S., Norris, K. & Kemp, N. (2017, November). *Cognitive outcomes and intervention attendance of Tasmanian preterm preschoolers.* Poster presentation at the Allied Health Symposium, Hobart, Australia.

Cognitive outcomes and intervention attendance of Tasmanian preterm preschoolers

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Background: Children born preterm are at a higher risk of having developmental and cognitive difficulties[1]. Social and medical risks can have negative impacts on outcomes[2].

Aim: To investigate cognitive outcomes, attendance rates in therapy and associated social and medical risks between preterm preschool children from South (S) North (N) and North-West (NW) Tasmania.

Participants: 141 children from the Royal Hobart Hospital (RHH) preterm infant follow-up program born in 2007-2009 (70 males, 71 females; gestational age 23.6-32.5 weeks, *M*= 29.6 weeks). 48% from South (S; *n*=67), 28% from North (N; *n*=39) and 24% North-West (NW; *n*=35) Tasmania.

Method: one-way ANOVA and $\chi 2$ test were used to compare the S/N/NW regions on the below variables

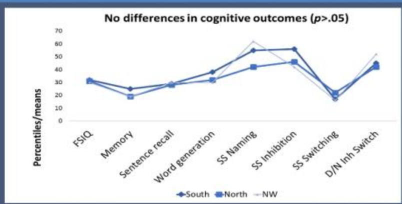
Assessment at 4-years

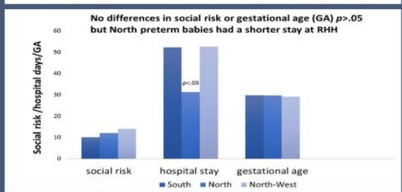
- -WPPSI-III
- -NEPSY-II: Narrative memory, Sentence recall, Word generation
- -Day-Night Stroop (D/N)
- -Shape School (SS Naming, Inhibition and Switching)

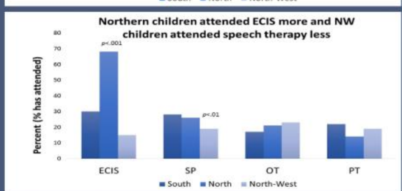
Risks & intervention attendance measures

- -Social risk scale
- -Medical risk: length of stay at RHH
- -Gestational age
- -Attendance at the Early Childhood Intervention Services (ECIS), speech therapy (SP), occupational therapy (OT) and physiotherapy (PT) at 0-4 years (current or previous)

Results: There were no significant differences in developmental or cognitive outcomes between the preterm preschoolers from South, North and North-West Tasmania (p>.05).
The social and medical risk factors were similar (p>.05, apart from a shorter stay at the Royal Hobart Hospital for the children from N Tasmania when compared to S and NW (p=.001). There were differences in attendance rates in early intervention and speech therapy (combined current and previous attendance). ECIS attendance of children from N was highest (p<.001), and the children in NW had attended speech therapy less than children from S and N (p<.01). There were no other differences in attendance in therapies when prior attendance was included, although the "current" attendance rates in OT and PT also varied.







Conclusions: Tasmanian preterm preschool children did not have significant differences in risk factors or developmental and cognitive outcomes based on their residential region, albeit their intervention attendance rates for ECIS and SP varied. The shorter length of stay in hospital for children from North may be explained by transfers to the neonatal unit at the Launceston General Hospital. It could be useful to investigate if intervention attendance differences were due to the factors associated with the preterm children or service delivery. Also, further studies investigating associations between educational/allied health intervention and cognitive, developmental and academic outcomes of preterm children are warranted.

This study has been supported by grants from the RHH Research Foundation

ROYAL HOBART HOSPITAL RESEARCH FOUNDATION

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[1] Anderson, P. J., & Doyle, L. W. (2003). Executive functioning in school-aged children who were born very preterm or with extremely low birth weight in the 1990s. *Pediatrics, 114*, 50–57.
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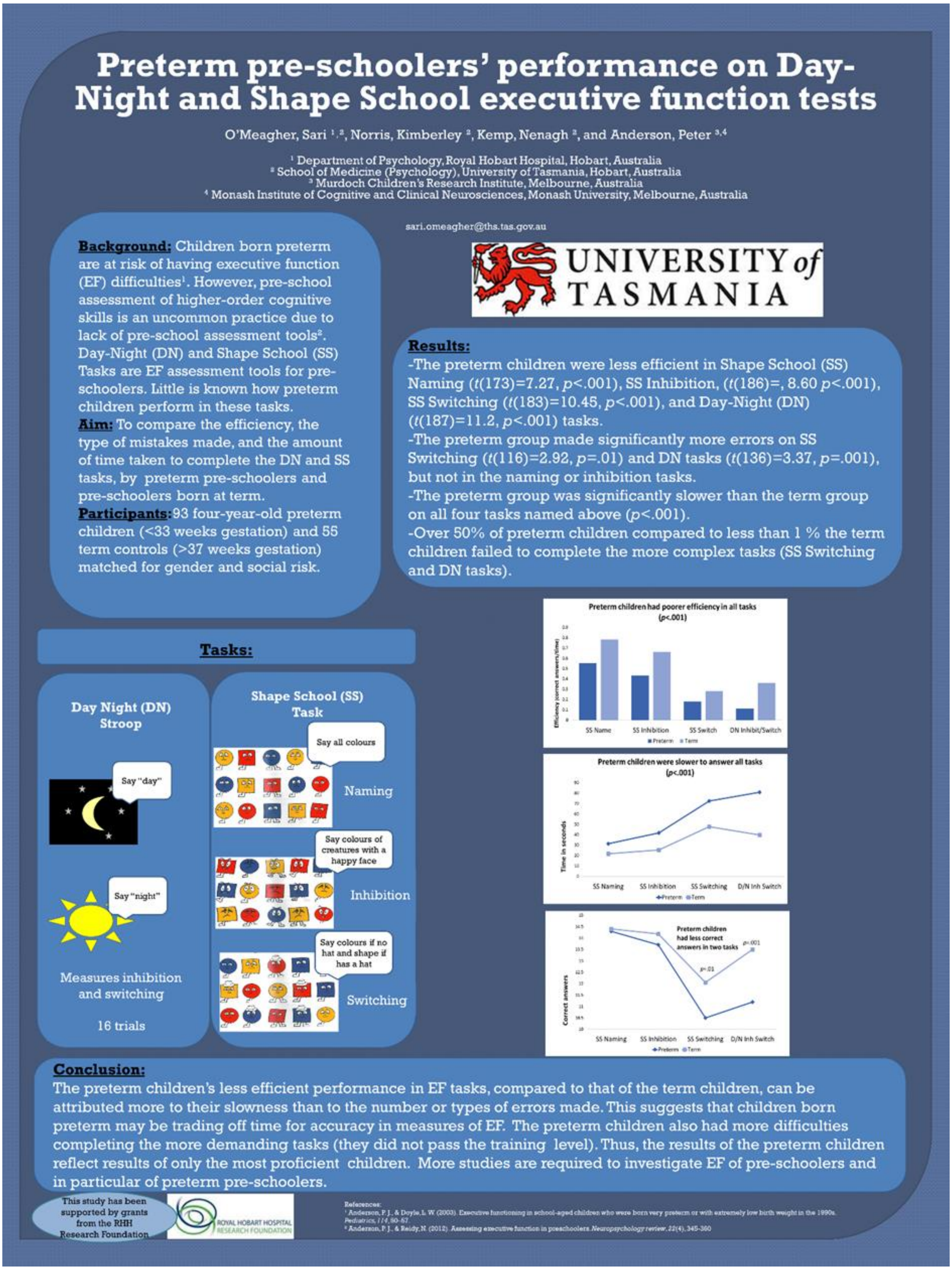
Appendix C. O'Meagher, S., Kemp, N., Norris, K., & Anderson, P. (2017, September). *Preterm preschoolers' performance on Day-Night and Shape School executive function tasks.* Poster presentation at the 6th Scientific Conference of the Federation of the European Societies of Neuropsychology (FESN), Maastricht, the Netherlands.



Appendix D. O'Meagher, S., Kemp, N., Skilbeck, C. & Anderson, P. (2015, July). *Does the performance of preterm preschool children on executive function tests predict executive functioning in real life?*. Poster presentation at the 5th International Neuropsychological Society Pacific Rim Conference, Sydney, Australia.

Does the performance of preterm preschool children on executive function tests predict executive functioning in real life?

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Background: School-age children born preterm are at risk of developing executive function (EF) difficulties[1]. However, preschool assessment of higher-order cognitive skills is an uncommon practice[2], and relatively little is known about how to identify children most at risk of developing EF difficulties prior to entering school.

Aim: To investigate the usefulness of current psychological assessment tools in predicting executive functioning in everyday life in preterm preschoolers.

Participants: 127 children from the Royal Hobart Hospital preterm infant follow-up program born in 2007-2009(61 males, 66 females; gestational age 23.6-32.5weeks, *M*= 29.6 weeks).

Procedure:

Assessment at 4-years

-WPPSI-III
-NEPSY-II: Narrative memory recall, Sentence recall and Word generation
-Day-Night Stroop
-Shape School

Teacher and parent BRIEF-P questionnaires next year

-rating children's EF abilities in everyday life

-75 % of the parent and 95% of the teacher ones were returned

Results:
-Low verbal IQ at preschool predicted teacher-reported self-control, flexibility and emergent metacognitive (working memory and planning/organizational skills) problems at kindergarten.
-Low performance IQ predicted parent-reported self-control and emergent metacognitive difficulties.
-Efficiency in the Day-Night and Shape School tasks predicted only emergent metacognition.
-Word generation predicted parent-reported self-control, cognitive flexibility and emergent metacognition and teacher-reported emergent metacognition.

Test results predicting questionnaires

Parent BRIEF-P questionnaires

Test results	Self-Control		Flexibility		Emergent Metacognitive	
	b	β	b	β	b	β
Verbal IQ	-0.04	-0.14	-0.10	-0.40	-0.08	-0.22
Performance IQ	-0.18***	-0.43***	-.006	-.18	-0.19***	-0.40***
Narrative Memory Recall	0.02	0.03	0.01	0.02	-0.03	-0.03
Sentence Recall	-0.07	-0.03	-0.01	-0.3	-0.07	-0.16
Word Generation	-0.12**	-0.32**	-0.12***	-0.42***	-0.17***	-0.41***
Day Night efficiency	-1.91	-0.02	-17.40	-0.25	-28.15*	-0.27*
Shape School A Naming	-5.07	-0.01	-6.90	-0.20	-17.46**	-0.35**
Shape School B Inhibition	-7.66	-0.16	-7.03	-0.19	-20.12**	-0.37**
Shape School C Shifting	14.28	0.13	-16.55	-0.20	-48.39**	-0.40**

Teacher BRIEF-P questionnaires

Test results	Self-Control		Flexibility		Emergent Metacognitive	
	b	β	b	β	b	β
Verbal IQ	-0.10***	-0.34***	-0.06***	-0.34***	-0.17***	-0.46***
Performance IQ	-0.03	-0.08	0.00	0.02	-0.08	-0.17
Narrative Memory Recall	-0.02	-0.03	0.00	-0.01	0.12	0.13
Sentence Recall	0.03	0.08	0.04	0.19	-0.05	-0.13
Word Generation	-0.07	-0.21	-0.03	-0.16	-0.17***	-0.37***
Day Night efficiency	-11.38	-0.14	-10.45	-0.20	-31.33**	-0.30**
Shape School A Naming	-7.45	-0.19	-0.85	-0.03	-13.78*	-0.27*
Shape School B Inhibition	-4.60	-0.11	-1.39	-0.05	-11.69*	-0.21*
Shape School C Shifting	-7.83	-0.08	-6.58	-0.11	-33.73*	-0.27*

* $p \leq .05$
** $p \leq .01$
*** $p \leq .001$

Conclusions:
Preschool preterm children's results on intelligence and performance-based EF tests at 4 years had some predictive value for real-life EF difficulties, as reported by the parents and the teachers, when entering school.

There is a need for ecologically valid EF screening tools in order to better identify preterm preschoolers who may experience real-life EF difficulties at early school-age.

This study has been supported by grants from the RHH Research Foundation

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